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ABSTRACT

The conference reported in this document utilized a conceptual matrix to organize presentations on research priorities in prevention, definitive treatment, and ongoing management of impairment, disability, and handicap. Ten research strategy recommendations developed at the conference are offered for implementation by The Easter Seal Research Institute in Ontario. Authors and titles of major papers included are: "The Role of Epidemiological Studies in Neuropediatric Handicap Research" (Lennart von Wendt); "The Role of Epidemiology in the Study of Physical Disability in Childhood" (Gregory Sherman); "Prevention--The Molecular Level" (Nicholas Lenn); "Priorities in Prevention of Cerebral Palsy" (Graham Chance); "Reduction of Severity" (Martha Piper); "Reduction of Disability--Communication Technology" (Alan Newell); "Priorities for Research in the Field of Augmentative Communication" (Janice Light); "Functional Improvement--Environmental Control & Mobility" (Stephen Naumann); "Reduction in Disability--Advances in Technology" (Morris Milner); "The Psychosocial Impact of Handicap on Children and Youth" (Dennis Harper); "Research Priorities in Cerebral Palsy" (Lewis Rosenbloom); "Spinal Dysraphic States" (John Banta); "Education and Research Training" (Carol Richards); and "The Use of Correct Scientific Measurement and Analysis" (David Winter). (JDD)

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THE EASTER SEAL RESEARCH INSTITUTE

RESEARCH PRIORITIES IN CHILDHOOD DISABILITY

CONFERENCE PROCEEDINGS

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Easter Seals

UNIVERSITY OF WESTERN ONTARIO, LONDON, ONTARIO, CANADA

PRIORITIES FOR RESEARCH IN CHILDHOOD PHYSICAL DISABILITY

10th - 13th May, 1988

Funded by

THE EASTER SEAL RESEARCH INSTITUTE (ONTARIO)

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Professor Lennart von Wendt is sponsored by

THE HOSPITAL FOR SICK CHILDREN FOUNDATION

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Foreword

The decision to publish the proceedings of an international conference is easily taken. Keeping that promise is difficult. The publication of this volume involved a small group of people in a great deal of exhausting and time consuming work. The major task of assembling and editing the individual papers, initially prepared for oral presentation, was undertaken by Dr. A. Mervyn Fox, Chairman of the Conference Planning Committee.

Since the original papers, presented to the Conference, had different type-faces and layouts, in order to make a readable publication, a standardized format was required. Our energetic Secretary, Dr. Cyril Greenland, was responsible for coordinating this production.

As President of The Easter Seal Research Institute, it is my privilege to thank the distinguished group of clinical scientists who have allowed us to publish their excellent papers. We are also indebted to those involved in the production and distribution of this volume. However, for reasons beyond our control, it was impossible to include transcriptions of the informal discussions which enlivened this unique Research Priorities Conference.

The cost of publishing and mailing these proceedings was supported by grants from the Health Services and Promotion Branch of Health and Welfare Canada, and the Policy Development and Research Division of the Ontario Ministry of Health. The generous support of these bodies is acknowledged with gratitude. Dave Waters, Director of Finance, Thames Valley Children's Centre, was most resourceful in managing the Conference funds.

Readers of this report will, no doubt, be interested to know that our Grant Review Committee and Board have accepted in principle the Conference recommendations presented by Dr. Fox in chapter 2, page (vi). In order to fulfill this mandate, the role of The Easter Seal Research Institute must change. Difficult though it is, we will try to move beyond providing researchers with seed money. Instead, in order to enable interdisciplinary teams to dedicate themselves to well defined research projects for periods of between five and ten years, our financial strategies will be revised. At a time of economic restraint, this will take some years to achieve, but the first steps have already been taken. The funding of innovative, multi-disciplinary research, dedicated to the prevention and treatment of childhood disabilities, has never been more important and the prospects of success so promising.

F. Howard Keast

F. Howard Keast
President
The Easter Seal Research Institute

2. Introduction and recommendations

A. Mervyn Fox, M.B., B.S., F.R.C.P (C), D.C.H.
Chairman,
Conference Planning Committee

The Conference held on the campus of the University of Western Ontario in May 1988, funded by the Easter Seal Research Institute, the Thames Valley Children's Centre, and the University Department of Paediatrics, with further support from Provincial and Federal Government sources, was a very considerable success in its own right. The evaluations were uniformly highly positive. The Conference remains alive in the memories of those who attended as a unique opportunity for outstanding minds from many different academic disciplines, united only by their intellectual rigors and their commitment to the physically disabled, to commingle and to stimulate each other in discussion. The Conference was relatively unstructured, allowing as much time for discussion and debate as for formal presentation, and it was this feature which drew most praise from both invited and paying participants.

The Conference was planned utilising a conceptual matrix developed from the World Health Organisation definitions of Impairment, Disability and Handicap with sessions related to prevention, definitive treatment and on-going management in each category. Outstanding researchers were invited to prepare presentations on research priorities in each cell of the matrix, and these papers, which are submitted with this summary report, were pre-circulated to distinguished discussants whose responses ranged from elaborate further documents to more informal oral commentaries. After the planned contributions, at least half of each session was dedicated to free debate. Whilst the invited Faculty ably reviewed specific priorities within their fields it was not expected that some cookbook recipe for the conduct of research would ensue. The Conference planners were hoping rather to establish a philosophical consensus on the way in which research funding in the Province might best be deployed for the widest benefit to the young physically disabled population. No specific research project emerged as demanding immediate attention, nor was such conclusion anticipated. The constituency subsumed as "physically disabled" is very heterogeneous and has somewhat blurred boundaries. Whilst cerebral palsy, itself a poorly defined and little understood clinical entity, was often adopted as a model for the opportunity of prevention and the delivery of rehabilitation services, the Conference was repeatedly reminded that this condition accounts for only 20% of the caseload of The Easter Seal Society. Rehabilitation professionals, both clinicians and researchers, tend to be more impressed by the commonalities - the shared social problems, the problems in obtaining helping care, the absence of truly validated interventions - between widely disparate groups.

Whilst all the Principal Speakers reviewed state-of-the-art knowledge in their areas, they tended to emphasise, properly, the vastness of our ignorance and the imperative need for the expansion of what is known. Of particular concern to The Easter Seal Society must be the consensus, apparent in many discussions, that our intervention programs and therapies are almost entirely untested and that much that we do is supported by faith rather than by science. Methodology exists for validating most, if not all, programs aimed at reducing the incidence of impairment, for alleviating the severity of disability and for obviating handicap, and the application of funding to appropriately conducted research is a priority for the entire rehabilitation community - professionals, parents, volunteers and children - exceeded only by the paramount necessity of ensuring that the results of such research are in fact implemented. One of the most outspoken and influential participants in the Conference, on a philosophical level, was Professor Alan Newell of the Microcomputer Centre, University of Dundee, Scotland. It is relevant to quote from his address some of the ideas which molded the ultimate consensus:

"The production of new ideas cannot be turned on like a tap, nor can a single mind generate the new ideas which should be generated. The most useful action which can be taken, however, is to create an infrastructure and atmosphere which encourages new ideas.The strategy one adopts critically depends upon the time-frame in which a solution is expected. Interaction between people working in different time-frames is essential, but equally essential is that each worker should know what his or her primary time-frame is. A research strategy with a 5- to 10-year time-frame should not be too closely focussed upon today's perception of clinical needs,which are often articulated as the detailed inadequacies of current systems."

(Professor Newell was talking of computer systems in the field of augmentative communication, but his statement and theme apply just as well to the health care and rehabilitation delivery systems, which are conspicuous in their failure to implement what is already known.)

"Service functions should be linked to research environments to their mutual benefit.

Because the needs of the two environments can be very different, however, it is counter-productive for short-term needs to be allowed to dominate a research environment. In a five to ten year time-frame:

(i) The primary interest should be in radical change, the effects of which cannot be fully foreseen.

(ii) Good ideas are unlikely to come entirely from within one discipline. They will break new ground and be contrary to received wisdom.

(iii) Unless an idea creates at least some scepticism, or even hostility, it is unlikely to be really novel.

(iv) It is difficult to predict losers and impossible to predict winners in these situations.

We need to encourage a wide perspective in our researchers - clearly a thorough knowledge of current practice - but also a healthy scepticism, and we must be prepared to back their hunches. Funding agencies should be wary of being too conservative and only supporting work which will clearly be successful. The future is a gamble and the longer the time scale the longer the odds."

One of the consensus recommendations is for the creation of research teams at Centres of Excellence and here again it is apposite to quote Professor Newell.

"The research team should be fully familiar with but not dominated by service needs. Researchers should spend a greater proportion of their time thinking about what they want to achieve rather than how they want to achieve it.The team should be fully multidisciplinary and advantages would accrue from including representatives from unfamiliar disciplines".

One of the underlying themes at Conference discussions was the frightening extent to which Rehabilitation Science is displaced by Rehabilitation faith, empiricism, and tradition, and the urgent need to train and put in place both as clinicians and researchers individuals capable of bringing critical and radical reappraisal to everyday practice and received wisdom. This deficit is inevitably most extensive in the alleviation of disability and handicap. The proper concern of The Easter Seal Society, rather than in regard to current research and practice is the basic sciences relevant to the understanding and primary prevention of Impairment, or to the clinical applied sciences of surgery and pharmacology whose efforts may also be directed to modification of the fundamental Impairment level. The requirement that future funding continue and elaborate the present Easter Seal Research Institute standards of adherence to intellectually rigorous methodology and high-level peer-review process was implicit, together with the underlying agenda of shifting, not only research and program evaluation but also clinical practice in the direction of rational science.

A supplementary theme to be reflected in the final recommendations was the importance, in all areas, of a sound epidemiological data-base, without which it is impossible truly to priorize research needs, or to conduct much of the research that is truly needed. Absence of an accurate epidemiological data-base relevant to the physically disabled of Ontario impacts not only upon research strategy and execution, but upon manpower planning, public financing and administration of health, social and rehabilitation services. Specifically, without such a data-base, it may prove impossible to conduct much needed research upon rare diseases, to study sub-types and natural outcomes of common impairments, or to evaluate the impact of social change, education, or rehabilitative effort.

With these preliminary remarks of a philosophical nature - for this was if anything a philosophical conference - and recalling that many excellent tactical or project-related statements of priorities are contained both in the primary papers presented here let us proceed to list consensus recommendations for research strategies of The Easter Seal Research Institute.

Recommendations

1. ESRI should, probably in collaboration with one or more University Departments and preferably with the support of the Provincial Government, establish prospective Registers relevant to the impairments and disabilities within its sphere of interest, which should include the communication disorders. Such Registers are not to be construed as so-called "at-risk Registers" of potential as well as established instances of vaguely defined entities of disadvantage or disablement, which have proved unmanageable and without benefit or scientific basis. Registers should be constructed to facilitate the identification of cases of, and hence the study of, rarer disabling impairments, and the definition of sub-types of the established major disabling conditions. Registers would provide a prospective but flexible data-base on a provincial level, accessible

freely within the limits of confidentiality to approved researchers on an inter-agency basis. The commitment to creation and maintenance of such Registers would likely require a specific endowment because of the essential long-term commitment. Such epidemiological Registers require full-time directors or, if small, a major commitment from appropriate epidemiological and clinical experts. ESRI would make an outstanding contribution to the physically disabled in the province by the establishment of such Registries, but would also provide a powerful role-model for research and practice in other areas nationally and internationally.

2. ESRI should facilitate the establishment of Centres of Excellence for research into habilitation and rehabilitation science related primarily to prevention and management of Disability and handicap. Centres of Excellence would utilise existing University Departments and Treatment Centres. Funding would not be project-based, but would be directed to the attraction of interdisciplinary research and teaching staff and to the creation of an appropriate research environment with a loosely defined but closely monitored mandate and sphere of interest. Support should be extended for a 5- to 10-year period. Centres of Excellence might be closely associated with the maintenance and exploitation of specific research Registries.

3. ESRI should facilitate the networking of existing University Departments and Treatment Centres by supporting record-linkages and data-exchanges, information about work-in-progress and areas of expertise, and by sponsoring authoritative conferences on specific topics.

4. ESRI should continue its present emphasis on the training of research personnel.The greatest need would appear to be for post-doctoral training in the rehabilitation professions, for it is by the creation of a cadre of teachers with extensive research experience that the greatest multiplier effect on the education and practice of rehabilitation professionals, and on research into specific fields of physical disability, will be achieved. However, it is not generally realised how few teachers in the rehabilitation sciences have any significant research experience or how this influences the balance of faith and science in curriculum; and there is ample justification for some funding for research training below the post-doctoral level and for continuation of the present program of undergraduate scholars.

5. ESRI should declare a priority interest in the validation of established treatment systems as well as in the evaluation of innovative rehabilitation programs and devices. ESRI should facilitate the linkage of the Provincial Treatment Centres in such activities.

6. While there was no consensus that ESRI should not fund basic scientific research, or primarily medical or surgical clinical research into prevention and treatment of the impairments causing physical disability, it was recognized that such areas have their own research funding sources. In contrast research into rehabilitation, disability and handicap is even even more underfunded and disproportionately dependent upon ESRI for its promotion. Rehabilitation Science, therefore, should be the recognised priority area for ESRI project support.

7. ESRI should develop financial strategies appropriate to a policy of funding long-term research projects, so distinguishing itself from current practice among granting agencies. Physical disability is always a developmental process, the result of permanent impairments creating changing disability and social disadvantage as an individual advances in years. Results of interventions must be studied both immediately and over time if their true impact is to be understood. Many interventions are themselves prolonged. Establishment of epidemiological registries will serve this end up to a point, but priority should also be given to specific longitudinal outcome or natural history projects.

8. ESRI should, possibly in collaboration with Provincial Government and appropriate University Departments or Treatment Centres, undertake on an interdisciplinary basis province-wide studies of the health care delivery system. This should focus upon health and rehabilitation services for the physically disabled in terms of their needs, financing, structure, effectiveness and efficiency. The results of such research should be fed back into the system in an authoritative manner such that changes and improvements in existing organisations and practices can be rapidly implemented. Utilisation of existing governmental data-bases,such as that associated with ADP, should be explored. ESRI could justifiably assume a prime role in evaluation of service delivery in terms of financing and systems organisation, rather than simply concentrating upon conventional micro-

program evaluation projects, essential as these are.

9. ESRI should acknowledge that today's young physically disabled become tomorrow's aged, with multiple unmet needs and a general ignorance and lack of appreciation of their unique problems, both on an everyday clinical level and in terms of research activity. It is suggested that, possibly in collaboration with existing adult-oriented service organisations, ESRI stimulate research into the natural history and clinical management or rehabilitation of the aging person with such developmental conditions as cerebral palsy, spinal dysraphic states, poliomyelitis, scoliosis and congenital amputations, cleft palate, and the orthopaedic malformations and deformities. Academic chairs dedicated to the adult mentally retarded are now being created in Ontario, and ESRI should consider the possibility of supporting such an endowment for academic study of the physical disabilities of childhood in adult life.

10. No firm consensus was reached as to the proper relative priority, in terms of disbursement of available funding, to the above priority areas, but overall it was felt permissible to continue funding of individual projects, particularly those concerned with rehabilitation science and achieving the highest standards in terms of methodology, to around 40% or 50% of whatever was available. Special funding or endowments might need to be created initially for the achievement of the more important strategic objectives outlined above, which should account for at least 50 to 60% of annual expenditure. Approximately equal proportions of this amount would be used for training of research personnel, establishment of specific registries, multi-agency networking and the creation of Centres of Excellence in research, and for the other strategic objectives. These figures are quoted less as explicit guide-lines than to emphasise the strong priority-weighting placed by the consensus upon the strategic objectives for ESRI as opposed to the tactical gains achieved by project funding, thus implying a radical change in ESRI financial policy and deployment.

A final consensus recommendation was that a similar Conference be held in approximately five years to review the impact of 1988 and to re-define priorities for the fast-approaching Twenty-first Century.

3. The role of epidemiological studies in neuropediatric handicap research

Professor Lennart von Wendt
Bracke Ostergard
Regional Pediatric Rehabilitation Centre
and Department of Handicap Research
University of Gothenburg, Sweden

Research of disabling neurological conditions in the paediatric population has traditionally been conducted along several main pathways.

New clinical entities and syndromes are still being described. Such scientific contributions have frequently been considered superficial and descriptive. These contributions, however, represent a quite necessary first step in the process of sorting out combinations of symptoms and signs into diagnoses.

A very good example of this process is the history of Rett's syndrome, originally outlined by the Austrian Andreas Rett in 1966 (12). The significance of this identification of a previously unknown clinical entity remained obscured until the 80's. Today this disorder is considered as one of the most important neurometabolical causes for severe mental retardation among females. At present well over 1,000 patients have been found, and the prevalence of the classical form in Sweden is around 1:10,000 girls (62).

Once a new syndrome has been established, it is time to proceed to the next step. This consists of reports describing the pathogenesis and underlying biochemical defect or mechanism of maldevelopment. Especially successful studies have resulted in creating a basis for therapy, such as replacement therapy in a number of handicapping metabolic disorders.

The approach outlined above represents typical clinical research, without any epidemiological features.

In epidemiological studies morbidity is usually analyzed from a different point of view, without focusing on clinical or biochemical details.

Epidemiological neuropediatric research already has, and also in the future should, contribute in the following ways:

1. Studies of the distribution of disease in cluding incidence, prevalence and secular trends.

2. Analysis of etiology and distribution of risk factors.

3. Long-term follow-up studies with respect to prognosis of the disorder and social conse quences for the patient and his family.

4. Analysis of the consequences of chronic neurological disease on health care, social welfare and educational service in society.

5. Analysis of the effect of changes of methods and organisation of health care upon morbidity.

Such data forms a necessary basis for decision concerning the allocation of social and health care resources. Especially when chronically handicapping conditions are concerned a thorough knowledge about care load and prognosis are essential for planning purposes.

Sources of epidemiological data relevant for neuropediatrics

Epidemiological data can be compiled in several ways. The type of data available is variable. The organisation of health care and legislation are the major causes for this variation.

1. Administrative registers and vital statistics: These are the ideal tool when the analyses require very high numbers of individuals. Analyses of the consequences of secular trends and development of the health care system should be performed on the basis of such information. Such sources are of very limited usefulness when analysing individual diseases, and especially rare diseases.

In countries and regions with a strict centralized and society-controlled health care organisation morbidity and contacts with the units of health care are usually registered. Examples of this are for instance nation-wide birth registers and hospital discharge registers. Such sources are available in the nordic countries. Since these individuals are relatively easily identifiable this data can be used for a variety of analytical approaches, which can be further augmented by retrieving information also on drug consumption, sick leave, employment etc.

The drawback of such registers is the fact that there is virtually no possibility to control the accuracy of the compiled information. The available information is usually relatively crude as it mainly consists only of diagnoses. In the study of Northern Finland birth cohort of 1966 such register-based information has been utilized (10). Fortunately, information was also collected in other ways as it soon became evident that a very erroneous panorama would have been created if the national registers had been accepted as the single source of information.

2. Registers of a specific disorder compiled by expert scientists:

This is the ideal material for the detailed analyses of clinical, etiological and other aspects of a single disease. Examples of such data are the Danish register of mental retardation, and the Danish, Swedish and Australian registers of cerebral palsy (4,5). The degree of ascertainment and the consistency of applied criteria for inclusion can be assumed to represent a very high standard in such data.

Types of design

The most popular design is the case-control-study. In neuropediatric research a widely used method implies the collection of cases and matched or unmatched controls. The matching is not infrequently a complicated procedure, and the ideal solution is depending on the central hypothesis of the study. Collecting controls so that all rules are obeyed is laborious and expensive. Ethical aspects can also create problems. A typical case-control-study is the Swedish CP-series, in which etiological risk factors have been analysed by means of carefully compiled healthy controls (4,5).

The cohort studies have been far less frequently used. There are some relatively well-known series of this type, which have contributed to neuropediatric knowledge (1,8,10). Usually children or pregnant women have been enrolled in these studies, and thereafter a huge amount of information has been collected prospectively. Examples of this are the English 1958 cohort comprising of some 15,000 children, the American NCPP study comprising of 55,000 children and the northern Finnish 1966 and 1985-86 cohorts(1,10,11).

This approach is at least theoretically the ideal one in many respects:

- the collection of data is prospective, or at least should be so
- the study population is clearly defined from the start
- the degree of ascertainment of appearing disorders is high
- there is a unique possibility to follow the same individual from pregnancy to senility

Naturally, also drawbacks exist:

- rare conditions appear with a low numerus, despite the huge amount of individuals originally recruited. Many neuropediatric conditions have a prevalence in the range of 1 per 1000 or even less. This means that even if a cohort of 100,000 children is at hand, it is very difficult to make any conclusions about etiology and other aspects related to very rare disorders on the basis of cohort studies.
- the prospective compilation of data is very laborious and also expensive
- from the viewpoint of the academic career, prospective cohort studies are far from ideal, as the really interesting results can be obtained only after an interval of many years. This fact, which also creates problems for fund raising, certainly is a major reason for the fact that large cohort studies are relatively rare.

The conclusion is that there is no ideal epidemiological material, but the type of study design must be chosen on the basis of the type of question one is trying to elucidate.

A case-control-study is the optimal design when various aspects of a relatively rare disease are studied. These studies are almost always retrospective in nature.

A cohort serves very well when the occurrence of etiological and risk factors for various types of suboptimal development and morbidity are analysed. It also gives very reliable information about the total panorama of morbidity and aberrant development in the group studied, which usually can be considered representative for the population from which the sample has been drawn. The longitudinal aspect is another asset.

Nordic epidemiological studies related to pediatric neurology - some recent results

The few examples which are presented here illustrate the more theoretical considerations presented above. Simultaneously, some important issues in the field of neuropediatric epidemiology are presented.

1. *Contribution of early neonatal care to the recent decline in early neonatal mortality rates in Norway (7).*

Rolv T. Lie and Lorentz M. Irgens
Paper presented at the XI Scientific Meeting of the International Epidemiological Association, Helsinki, August 1987.

Source of data: Medical Birth Registry of Norway

Patients

126,000 live firstborn singletons weighing 1000 grams or more, born 1967-1971 and 105,000 infants fulfilling the same criteria born 1980-1984.

Object of analysis

The decline of early neonatal mortality from 0.67 to 0.24 per cent: role of neonatal care vs. sociodemographic factors.

Results

88.7% of the improvement was attributable to medical improvement and 11.3% attributable to socio-demographic factors.

Birthweight	Medical Improvement %	Sociodemographic changes %	Sum %
1.000-1.499	29.7	7.3	37.0
1.500-1.999	19.6	3.9	23.5
2.000-2.499	12.0	2.4	14.4
2.500-3.499	16.1	0.8	16.9
> 3.499	11.3	-3.1	8.2
Total	**88.7**	**11.3**	**100.0**

Comments

Such an analysis requires a huge amount of material because of the statistical methods used. This study is a very good example of how a birth register can be used. The result once again shows a clear net gain of improved neo-natal care especially in low birth weight infants.

2. Prevalence of cerebral palsy in Sweden in children born 1959-1982. The 4-year period 1979-1982 and its relation to the 20-year-birth period 1959-1979 (4).

Hagberg B, Hagberg G, Olow I, v. Wendt L.
Department of Paediatrics II, University of Gothenburg and Bracke Ostergard Regional Pediatric Rehabilitation Centre, Gothenburg, Sweden. Manuscript 1988.

Source of data: Register of children with CP in the south-western Swedish health care region (Gothenburg area). Population 1.9 million.

The series has, during the period, been compiled by the same authors according to uniform criteria.

Results

Birth year period	Prevalence per 1,000 liveborns
1959-62	1.9
1963-66	1.7
1967-70	1.4
1971-74	1.6
1975-78	2.0
1979-82	2.2

The prevalence in term children (1.1 per 1000) remained essentially stable until the period 1971-1974, but rose to 1.3 and 1.2 respectively in the two last periods. The variation is thus essentially attributable to changes in the preterm group. The lowest prevalence, 0.35 per 1000 was encountered in the period 1967-70, and the highest, 1.0 per thousand in the last period. Within the subgroups the most conspicuous variation was noted among the preterm diplegic infants.

It is essential to note that the rise of prevalence which first was noted in the period 1971-74 continues to rise. Especially alarming is the fact that the proportion of CP children with major additional handicaps has increased and this phenomenon is further pronounced in the last period.

This, not too encouraging finding should be viewed against the fact that the perinatal mortality in the period 1979-1982 was 7.2 per thousand, which was 3.2 less than in the preceding period. As the increase of CP was 0.14, there still is a major net gain of healthy infants. However, a possible increase of other handicapping conditions, such as MR, infantile hydrocephalus and epilepsy have not been taken into account.

The findings of this study are in agreement with a number of other studies, in which this problem has been attacked from other points of view. The exact figures for prevalence of cerebral palsy should not uncritically be compared with those of other studies in other areas, as the delineation of cerebral palsy appears to vary. This variability continues to cause confusion and incomparable conflicting results of surveys.

Most generally accepted definitions of CP are based on the Little Club of London declaration "Cerebral palsy is a persistent, but not unchanging disorder of movement and posture appearing in the early years of life and due to a nonprogressive disorder of the brain, the result of interference during its development".

This definition has created several problems:

- early life has been very variably interpreted, and some authors separate the entity of late-onset CP, secondary to CNS infections and trauma, whereas others do not consider these conditions as CP at all.
- the definition represents a mixture of etiology and clinical characteristics.

The consequences of this definition and the existing variable sub-grouping are widely described by several authors in the volume **Epidemiology of the cerebral palsies**, and will not be covered in detail here. A general agreement on the detailed criteria and on the principles for the sub-grouping of CP would increase the international understanding in the field of CP research. It should, however, be concluded that at present the only reliable way of compiling longitudinal and reliable information about the prevalence of cerebral palsy is represented by the approach of the Hagberg group.

3. Cerebral palsy and other neuroimpairments in Sweden.

Bengt Hagberg and coworkers.

In addition to the well-known series "The changing panorama of cerebral palsy in Sweden", the Hagberg group has also carried out detailed analysis of aetiology, nosology and disability related to subtypes of CP (4). The most recent studies deal with spastic hemiplegia (Uvebarndt 1988) (14) and spastic tetraplegia (2). In the latter series, which have been collected by Karin Edebol-Tysk the study area has been expanded to cover a major part of Sweden. In this study special attention has been paid to care-load and parental coping.

In another study Elisabeth Fernell collected a series of infantile hydrocephalus, demonstrating an increase of the incidence among premature infants (3). This is another consequence of the increased survival rate of preterms. The most alarming finding is the observation of an increase of very preterm infants with infantile hydrocephalus in combination with other severe neuroimpairments.

When all these studies are considered together, and as similar population-based studies of epilepsy and mental retardation also have been carried out a very distinct picture of major neurological morbidity in Swedish children emerges. Some of these observations have prompted the Swedish Medical Board to issue recommendations concerning consumption of moderate intake of alcohol and tranquillizers during pregnancy, as these population-based studies identified alcohol as a major risk factor for mild mental retardation in urban areas and benzodiazepines as a teratogenic agent.

4. Prevalence of handicapping neurological conditions in the Northern Finland birth cohort of 1966 (10,11,15).

Rantakallio P. v. Wendt L.

Departments of Public Health Science and Pediatrics, University of Oulu, Finland.

Source of data: The 1966 one-year birth cohort of northern Finland.

12,000 pregnancies, 96% of all live-births in the year 1966, in the two northernmost counties of Finland were enrolled in the study. Data on socio-demographic factors, pregnancy, delivery, development and morbidity of the children as well as occupational training and entrance to adult life has been collected prospectively.

A similar birth-cohort comprising 10,000 live-births was in an analogous way collected in 1985-86.

Results

Up to the age of 16 years a total of 549 children (327 boys and 222 girls); 46 per 1000 had developed a disabling neurological condition (CP, Epilepsy, mental retardation or mental subnormality, congenital CNS malformations, CNS-trauma resulting in chronic disablement, hearing and visual defects). In a number of analyses, the role of social, obstetrical and perinatal risk-factors have been analysed.

Comments

Such a longitudinal study creates unusual possibilities for the assessment of the role of a huge number of factors for the long-term developmental and social prognosis. As such factors-prospectively have been entered the compiled information bank both for diseased and healthy children, there is a unique possibility to analyse the impact of various factors.

The new cohort, collected 20 years later than the first one creates an unusual opportunity to analyse the consequences of the development of antenatal and perinatal care on the other side, and the influence of changes in demographic factors upon morbidity and development. Preliminary results indicate that active intervention in pregnancies, identified in early pregnancy as being high-risk ones markedly reduced perinatal mortality and morbidity, as this analysis was carried as a case-control study.

The role of epidemiological research in the future

Within the field of neuropediatrics there still are many questions which have not fully been answered. In addition, the development of therapeutic methods appear to create new questions.

In the field of **pediatric neurology** at least the following questions are of special interest:

Mental retardation

At present, major confusion concerning a possible secular trend in prevalence of various types of MR seems to be the case. One could presuppose that improved maternal care, intrauterine or early detection of congenital malformations and metabolical disorders would lead to a reduction of at least MR caused by such factors. On the other hand, it seems apparent that only a relatively small part of the conditions causing especially SMR can be recognized early or in utero.

There is also indirect evidence for the fact that the increased survival rate of very low birth weight infants lead to an increase of children with multiple handicaps.

In addition, an increased survival rate of children with multiple handicaps and malformations may cause an increase of the prevalence. Therefore, it seems possible that the improvement of intrauterine detection of anomalies operates favourably on the individual level without major consequences for society.

A major methodological problem related to research on mental retardation is the definition. The delineation of MR is very much influenced by the society's attitudes towards the mentally retarded, and therefore figures of prevalence from different countries may not be comparable. At least a part of the secular changes may in fact reflect changes of attitudes and school systems.

Very important questions related to these matters can be resolved only by epidemiological studies. The same applies also to the educational and social consequences of mental retardation, with special reference to the principle of mainstreaming.

Continuous monitoring of possible changes in the panorama of neurological disablement.

This should be accomplished systematically in all countries for the purpose of:

- spotting untoward tendencies in time
- adjusting the allocation of resources

At present such studies are not systematically carried out. The necessary data could with relative ease be collected by means of registers of neurological disorders. This is, however, clearly controversial as the legislation in most countries seem to discourage large registers.

Very low birth weight infants

This group is a major concern in contemporary pediatrics. A better understanding of the pathogenesis of CNS damage characteristic for these individuals is emerging. At present, relatively little can be done to normalize and stabilize cerebral blood flow, which apparently is the crucial point. The results of perinatal care should be carefully monitored in a population-based way. This should be accomplished on a regional level in order to create appropriate feedback to peri- and neonatalogy.

Effects of new methods and technology

New therapies should never be accepted without proper randomized scrutiny. Unfortunately, medical practice still mainly is based upon tradition and "clinical experience". This statement is apparently also applicable to peri- and neonatal care. Analyses of long-term effects of various methods of management in the neonatal period are lacking.

An example of a therapeutic method which in the first place was introduced and accepted and only afterwards subjected to scientific studies is the use of intrauterine ultrasound for the detection of anomalies and disturbed intrauterine development. The impact of intrauterine ultrasound screening upon neurological morbidity remains to be analysed.

Early prognostic signs of neurological disablement.

Numerous attempts at constructing schemes for the systematic neurological evaluation of the small infant have been done. The predictability of most is so low that these tools are of very limited usefulness in clinical practice. Despite this, such methods are badly needed.

Social consequences of neurological disease

Such studies do exist, but a continuous follow-up is needed in order to monitor the effects of changes both in society and in the morbidity panorama..

Neuropediatric Rehabilitation

As compared to research in pediatric neurology, the discipline of pediatric rehabilitation as a branch of science appears very young and very weak. Most things are waiting to be done, and unfortunately this applies also to very basic questions.

Within pediatric rehabilitation, we usually differentiate between pediatric rehabilitation and pediatric habilitation. The former concept is applied for children and adolescents who acquire neuronal damage later in life, whereas habilitation deals with congenital or early acquired lesions. In the paediatric population the latter conditions dominate very clearly, accounting for over 90% of all disabled children, which is contrary to the situation among adults.

The history of neuropediatric re- and habilitation dates back to the post WW I era. The major groups are cerebral palsy, MMC and muscular disorders, especially Duchennes muscular dystrophy and mental retardation.

The pioneers in this field were usually not scientists in the classical meaning but innovators. The consequence is that the methods applied for instance in physiotherapy of cerebral palsy are not the results of systematic research work. On the contrary the methods used today are based on theories developed nonempirically some decades ago, by for instance the Bobath couple and Vojta and the Hungarian Peto.

There are consequently many reasons for epidemiologically oriented research in the field of neuropediatric rehabilitation:

The role of early started physiotherapy in children with a suspicion of CP.

There is a general agreement that such therapy should be started as early as ever possible, preferably in the neonatal period or at least during the first few months.

In some units or hospitals programs of risk-baby identification are being used, and infants who fulfill rather variable criteria of abnormal motor development are subjected to relatively intensive habilitation programs.

Several questions have remained unanswered:

A. At which age can a CP-like disorder safely be diagnosed?

B. Which criteria should be used for choosing the right infants for training?

C. Which children really benefit from these training programs?

D. Which children normalize spontaneously? Is it possible to identify those who heal spontaneously?

E. Are the therapeutic effects really due to therapy and not the spontaneous healing?

I think that these questions are extremely important since:

a. Involving the family in a training program has profound effects on the entire family for at least two generations.

b. An adequately performed training involves a major financial enterprise to be covered either by the family, insurance companies or society.

At present very few studies have addressed this question, all of them yielding very alarming results. On the basis of a risk-baby population Piper and coworkers (1988) (9) in a randomized trial failed to demonstrate any positive effect of an early training program.

Naturally the results are very difficult to interpret and it is almost impossible to account for confounding factors at least in a relatively small series.

I think that broad-based, by necessity multi-center studies of this important questions are badly needed.

Somehow, training programs for infants with delayed motor development are a classical example of the difficulties society runs into when a form of therapy is started without initial properly controlled studies.

Today, people working in this field fiercely oppose randomized trials claiming these to be unethical as clinical experience has proved the efficacy long ago.

The truth is, that every form of therapy which has not been subjected to such systematic control is unethical. I think that this should be emphasized especially when such large programs are concerned, as compared to single drugs, which usually have no effects on several generations.

Which training program is the best one?

If studies of the previous question are very rare, the situation is no better concerning this question.

The best recognized training programs are those of Bobath with a huge number of national modifications, Vojta, Peto and the very controversial Doman-Delicato method. None of these methods are based on the present concepts of neurophysiological functions.

One Swedish paper deals with a comparison between Vojta and Bobath-type therapy, failing to analyze the results as the starting point according to the Vojta system inevitably includes a number of completely healthy children which thus out of necessity would show brilliant results of therapy.

The prevailing confusion is by means diminished by the involved major commercial interests.

I think it is time to settle whether there are any differences between the therapeutic results of these programs. As CP is rare, this question also has to be dealt with within a large study. The multicenter approach appears a tempting solution.

Consequences of neurological handicapping conditions

In the paediatric age-group prognosis and effects upon family are relatively well known. However, these studies very rarely are population-based, which makes generalization of the results difficult.

Virtually nothing is known about the time when the child has developed into an adult.

Questions related to this can best be answered on the basis of a longitudinal study, but cross-sectional yields basic data about the present situation.

As some nations favour the principle of integration more clearly than other ones, the late effect of differing principles of the organisation of the care for these individuals could be analysed in the form of international collaboration.

General conclusions

In the field of neuropediatrics there is a very clear-cut role for the epidemiologically orientated research. Continuous monitoring of the panorama of morbidity in every region is mandatory. In this respect the south-west Swedish health care region serves as a model. Only population-based clinical data is firm enough to permit conclusions concerning organisation of health care. At least as important is the feedback which is generated in such a systematic monitoring. Unfortunately the total panorama of neurological morbidity does not become clearly visible until school-age, which inevitably delays the feedback.

The field of paediatric rehabilitation is, from a scientific point of view, an open field. Most questions, including basic epidemiological ones have been dealt with only to a very limited extent.

For a trained pediatric neurologist getting a proper schooling in the field of epidemiological methods may seem too laborious. However, neuropediatric epidemiology cannot be pursued without a good knowledge of this field.

The best practical solution implies collaboration between the pediatric neurologist and the epidemiologist and/or biostatistician. These can, however, not communicate in a meaningful way unless they learn basic facts from each other's professions.

References

1. Butler, N.R., Bonham, D.G., Perinatal mortality: The first report of the 1958 Perinatal mortality survey. E&S Livingstone,Edinburgh 1963.

2. Edebol-Tysk, K., Hagberg, B., Hagberg, G., Epidemiology of tetraplegic cerebral palsy in Sweden II. Prevalence, origin and birth data. (Manuscript 1988)

3. Fernell, E., Hagberg, B., Hagberg, G., v.Wendt, L., Epidemiology of Infantile Hydrocephalus in Sweden. I. Prevalence and General data. Acta Paediatr Scand 75: 9755-981, 1986.

4. Hagberg, B., Hagberg, G., Olow, I., von Wendt L., Prevalence of cerebral palsy in Sweden. The 4-year-year period 1979-82 and its relation to the 20-year-birth period 1959-1979. Manuscript 1988.

5. Hagberg, B.,Hagberg G., Olow, I., The changing panorama of cerebral ,palsy in Sweden. IV. Epidemiological trends 1959-1978. Acta Paediatr Scand 73: 433-440, 1984.6. Hagberg B, Witt-Engerstrom I: Rett Syndrome: epidemiology and Nosology - Progress in Knowledge 1986 - A Conference Communication. Brain Dev 1987:9.(in press)

7. Lie, R.T., Irgens, L.U., The contribution of early neonatal care to the recent decline in early neonatal mortality rates in Norway. International Epidemiological Association; XI Scientific Meeting, Helsinki, August 1987. Volume of Abstracts, p. 126.

8. Nelson, K.B., Ellenberg, J.H., The epidemiology of cerebral palsy. Adv Neurol 19: 421-35, 1978.

9. Piper, M., Mazer, B., Silver, K.M., Ramsay, M., Resolution of neurological symptoms in high-risk infants during the first two-years of life. dev Med Child Neur 30: 26-35, 1988.

10. Rantakallio, P., The longitudinal study of the Northern Finland birth cohort 1966. Paediatric and Perinatal Epidemiology 2:59-88, 1988.

11. Rantakallio, P., v.Wendt, L., Koivu, M., Prognosis of perinatal brain damage: A prospective study of a one-year birth cohort of 12000 children. Early Hum dev 15: 75-84, 1987.

12. Rett, A., Ueber ein cerebral-atrophisches Syndrom bei Hyperammonamie. Vienna: Brueder Hollinek, 1966.

13. Stanley, F., Alberman, E., The epidemiology of the cerebral palsies. SIMP, London 1984.

14. Uvebrandt, P., Hemiplegic cerebral palsy. Thesis, Gothenburg, 1988.

15. v.Wendt, L., Rantakallio, P., Makinen H: psychomotor development in the first year and mental retardation - a prospective study. J. Ment Defic Res. 28: 219-225, 1984.

Role of epidemiological research in neuropediatric handicap research

Discussion

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Dr. von Wendt has given us an excellent systematic presentation of the applicability of epidemiologic methods to research in physical disability. I agree with him that these methods have added much to our understanding in the field. Epidemiologic methods are applicable across the entire spectrum of chronic disorders embodied in the World Health Organization's (WHO) scheme. Explicit use of the WHO scheme should help focus the goals of epidemiologic research. I will cite a few examples of research questions, some of which have been actively pursued and some that remain to be refined and funded, that illustrate the application of epidemiologic approaches to aspects of childhood physical disabilities. These are intended to supplement the extensive list provided by Dr. von Wendt.

Diseases/Disorders

1. Epidemiology was founded on the investigation of the causes of disease. The successful identification and subsequent control of congenital rubella syndrome is an outstanding example of the strength of this approach. Quantitative epidemiologic methods will be major tools in identifying the apparent multifactorial causes of disorders such as arthrogryposis multiplex congenita.

2. The study of survival of populations of persons after disease onset and application of treatment owes much to epidemiology. Methods of survival data analysis have matured to the point where we should have rigorous expectations of ourselves when evaluating the efficacy of any new treatment intended to prolong life (Lee ET, 1980). Analytic techniques, life-table analysis, has been crucial to the success of some of the long term studies of survival of infants with myelodysplasia in our program (McLaughlin et al, 1985).

Impairments

Life-table analysis can be successfully applied to the course of individual impairments over time as easily as to survival of the entire organism. As applied to hip flexion contracture in myelodysplasia, we have been able to confirm the relationship to level of motor paralysis but have also seen patterns of change that suggest that there need to be important modifications of the textbook descriptions of lower extremity innervation (Shurtleff, 1986). The wider use of epidemiologic methods in this fashion should improve our understanding of individual impairments due to many causes of physical disability.

Disability

There have been many successful efforts to use the epidemiologic method for the study of long term disability. In addition to those cited by Dr. von Wendt there have been extensive studies in North America such as some of those forming the basis of the Vanderbilt Study (Hobbs et al, 1985). Dr. von Wendt also correctly identifies the study of the long term adult survival of persons with physical disability of childhood onset. Many of the on-going disputes of the management of mobility in children with cerebral palsy or spina bifida would resolve if we had reasonable feedback about long term consequences. There are many unknowns for adults as is exemplified by the recent recognition of long term deterioration post-polio. What are the consequences of the presence of a ventriculoperitoneal shunt or an artificial urinary sphincter? These are not trivial questions and the answers will depend on epidemiologic techniques in significant measure.

Handicap

While less clear intuitively, I believe that there is a role for epidemiologic methods in understanding the social process of handicap. The rigors of epidemiologic methods is beginning to find its way into opinion sampling and other domains in sociology and anthropology. We are beginning to benefit from the quantitative study of behaviour and social variables in the etiology of head injury and child abuse. We are beginning to understand how social handicap may be the cause rather than the result of physical disability.

Dr. von Wendt's presentation gave some well-known examples of good prospective cohort studies. While cohort studies are powerful and effective, there are some limits that must be respected to get the most mileage out of the time and cost involved. Dr. von Wendt calls attention to some of these limits. Classic cohort studies of chronic disease such as the National Collaborative Perinatal Project (NCPP) give excellent historical information, but if the field being studied is particularly dynamic, the results may be misleading in current application. In the case of the NCPP, many aspects of the obstetric care of newborns have changed in the last 20 years. The elimination of kernicterus as a cause of cerebral palsy is just one of many changes in the mix in the etiology of cerebral palsy than spans the duration of the NCPP. Similarly, the pessimistic outcome studies of the newborn management of spina bifida have been superseded by changes in treatment techniques. The approach utilized by the Swedish group using a time series of cohorts helps get around this problem by providing multiple intermediate points of reference from which one can estimate trends. We have found this approach useful in looking at survival in spina bifida (McLaughlin et al, 1985).

As Dr. von Wendt points out in his commentary on the Finnish birth cohort study, the quality and completeness of data collected and used in peidemiologic studies is often questionable. This is especially true if the data was collected for another purpose. Clinical data is notoriously unreliable. Most studies of the incidence of neural tube defects are badly confounded by the failure to distinguish between primary netural tube defects and secondary caudal cell mass abnormalities. Sometimes the definition of a critical variable is changed either by administrative fiat (as in the case of changes in the defintion of IQ cutoffs for mental retardation) or will differ from region to region (e.g. criteria differentiating stillborns and livebirths). Although harder to document, many clinicians talk about diagnostic "creep"; downward in the case of the definition of the limits of viable live birth; upward in the case of the scoring of IQ tests. Diagnostic creep occurs in the absence of formal changes in objective criteria and may be analogous to the gradual changes of connotation associated with the evolution of spoken language. The unreliability of clinical data can be improved by the use of standardized data collection with reproducibility and validity checks as quality assurance measures. Clinical groups in the United Kingdom (Evans et al, 1986) and Western Australia (Blair and Stanley, 1985) are working on these issues with cerebral palsy and the International Myelodysplasia Study Group is addressing the same problems in myelodysplasia (Shurtleff, 1986). Multiple source ascertainment is an approach to the usual problem of incomplete administrative records outside of countries with centralized clinical registries. All of this is dull and expensive to researchers and funding sources alike but must be confronted to get reliable results.

The cumbersome size of cohort studies can be modified by sample stratification based on characterization of variables thought most relevant to outcomes. Sophisticated statistical analysis is essential to such studies as Dr. von Wendt points out. Carefully measured multiple data points offset some of the need for size.

I am in complete agreement with Dr. von Wendt on the value of multicenter studies in the investigation of rare disorders. Multicenter studies may also be necessary to deal with the fine points of more common disorders as well. Such studies put great demands on researchers and funding sources. To amplify the return on such studies I believe that much more attention should be given to the training of primary investigators in techniques to manage such studies including budget control, personnel management, and critical path analysis. I also agree with Dr. von Wendt that there is little attention in current academia to doing long term cohort studies or obtaining the necessary skills to do them correctly. The malaise is tied partly to multiple demands on faculty, to the structure of publication/promotion cycles, and to the lack of long term funding commitments from support sources.

Epidemiologic research is not sufficient for studying all aspects of intervention to treat disability and impairment. Shorter term studies using two major types of experimental design approaches have a major place in this type of research. Classic prospective double-blind crossover studies as used with new medications are essential to the investigation of treatments for reasonably homogeneous groups. The statistical treatment of such studies are well described. Some problems such as mobility and communication intervention in cerebral palsy are so individual that the use of single-subject, repeated measures design becomes necessary. Basic work remains to be done on the generalization and statistical treatment of single-subject research but it is an important technique.

Finally, attention needs to be called to the importance of properly evaluating all methods of intervention for impairment and disability. Dr. von Wendt has properly called attention to the controversy over the use of physical therapy in cerebral palsy. His comments are generally applicable but physical therapists have no corner on the market for uncritical treatment. A wide variety of surgeries, medications, and other treatments have been employed over the years by a broad spectrum of health professionals with no critical evaluation of efficacy. The current wave of enthusiasm over selective dorsal rhizotomy will be yet another example if several well-designed studies are not launched in the next six to twelve months.

I hope this discussion has added some useful amplification to Dr. von Wendt's scholarly presentation. Epidemiologic techniques have broad application across the field of chronic physical disability in childhood and deserve the full support of public and private funding sources.

References

Lee, E.T., Statistical Methods for Survival Data Analysis, Lifetime Learning Publications, Belmont, Ca 1980.

McLaughlin, J.F., Shurtleff, D.B., Lamers, J.Y., et al. Influence of prognosis on decisions regarding the care of newborns with myelodysplasia, New England Journal of Medicine, 312:1589-94, 1985.

Shurtleff, D.B., et al, Myedolysplasia and Extrophies: Significance, Prevention and Treatment. Grune and Stratton, NY, 1986.

Hobbs, N., Perrin, J., and Ireys, H., Chronically Ill Children and Their Families. Jossey-Bass Publishers, San Francisco, 1985.

Evans, P., et al, Report of a meeting on the standardization of the recording and reporting of cerebral palsy. Developmental Medicine and Child Neurology, 28:543-549, 1986.

Blair, E., and Stanley, F., Interobserver agreement in the classification of cerebral palsy. Developmental Medicine and Child Neurology, 27:615-22, 1985.

4. The role of epidemiology in the study of physical disability in childhood

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As one who never comes face to face with a disabled child on a professional basis, I have been deeply impressed with the dedication and ingenuity of those who deal directly with this multi-faceted problem whether it is through physical therapy, biomedical engineering, orthopaedics, special education, or in some other way. However, I am also saddened that such efforts and activities are necessary. It would surely be better to prevent the occurrence of a disabling condition (in a primary sense) than to have to deal with it later in ways that are improving but will always be less than perfect.

Of all the major causes of physical disability in childhood, adverse reproductive outcomes are one of the most important. I will be concentrating today on diseases which produce disability as opposed to injury, unintentional and otherwise. For purposes of illustration, I will be using some information on the occurrence of spina bifida in Ontario.

Uses of epidemiology - The epidemiologic approach.

My presentation will of course be given from an epidemiological perspective. A popular definition of epidemiology is 'the study of the distribution and determinants of disease frequency in humans' (MacMahon and Pugh, 1970). Chronic disease epidemiology can provide essential information for:

1. identifying factors which increase or decrease the risk of disease or other health conditions and factors which influence the morbidity or mortality of affected persons

2. determining the relative importance of individual risk factors in the risk of diseases or health conditions caused by multiple factors

3. identifying high-risk populations

4. developing effective health care and prevention programs

5. evaluating existing health care and prevention programs to assess effectiveness.

The first three of these points would be familiar to most people. Thus an epidemiologist would approach the question of etiology in disease such as spina bifida by initially observing patterns and trends of the condition in human populations. Hypotheses would be formulated from such descriptive studies which could then be tested with a variety of pseudo-experimental ('analytical' in epidemiologic jargon) study designs.

The pace of epidemiology is often, by necessity, rather leisurely and seems to many outside the discipline to be maddeningly inconclusive. While it is probably true that a high tolerance for ambiguity comes in handy, it is also true that there is a general lack of understanding of the limits imposed by the ethical impossibility of using the artificial simplicity of animal experiments and of familiarity with the special techniques that have been and are continually being developed to deal with this special type of research.

Whatever the epidemiologist gives up in lack of experimental control is more than compensated for in the long run by the fact that results on humans are extrapolated to humans. Extrapolation of the experimentalist's results involve not only the difficulties of species differences, but also questions of equivalent dosages, of the relationship between the animal (or lower order) defect and the human defect and of the possible interactions of the exposure(s) with other factors in the human situation. Clearly, no single approach is 'better' or 'more scientific' than another; they are complimentary and should, if possible, be used in conjunction. Nevertheless, epidemiologic studies do have a distinct, practical advantage in 'complex' situations; i.e., those with necessary, multiple, simultaneous, low-level exposures which are interdependent and confounded with human lifestyle, occupational and other factors. 'Complex' situations are probably the rule rather than the exception in human chronic disease. In other words, if epidemiology is not well-suited to identifying precise causal pathways, it can be rather good at identifying intervention points which will break the causal chain. From the public health point of view it matters little where those points are.

The last two points dealing with developing and evaluating programs are less well appreciated and seldom carried out, at least in Canada. As the use of epidemiology in these areas grows, as it must do in a climate of strongly competing priorities and upwardly spiraling costs, basic systems of data collection and distribution must be improved across the country. Unfortunately, there are no coherent plans for a national system for the investigation and control of chronic diseases in Canada and no high-level realization that better basic data sources are needed. Apart from the etiology/risk factor issues, we are in the position, for almost all chronic diseases, of not knowing how many people are affected, where they are, what services they are using and how much those services cost, what services they might need but do not receive or whether the services they do receive are in fact cost-effective.

Current level of support

Having said that, the fact that very little is known about the causes of childhood disabling diseases cannot be too surprising. The situation with respect to congenital birth defects is typical (Fig. 1). In my opinion, the most important single reason for this is that reproductive health problems have received considerably less attention than cancer, respiratory and circulatory diseases. For example, in 1986-87, the National Health Research and Development Program (NHRDP) was supporting 15 projects related to the etiology, prevention or intervention of childhood disabling diseases. The NHRDP is the single largest source of funding for this type of research in Canada. The total expenditure over these two years was $443,784.05 (NHRDP, 1988), representing approximately 1% of the NHRDP annual budget or the cost of two three-bedroom bungalows in some Ottawa neighbourhoods. There is a bit of the chicken-egg problem here: does the lack of funding reflect a lack of interest or has a lack of money driven potential researchers to other areas? I don't know the answer to that question but I do know that advocacy - strident, persistent advocacy - gets attention. Such advocacy would not be misplaced; reproductive problems and their sequelae are of great public concern and are in fact associated with a tremendous emotional and public health cost.

In fact, the driving force today behind most issues is public perception. The impact of a worried and sincere public cannot be overestimated in creating public policy and hence public money. In general however, the public will not be worried until they are educated to be so and this takes time and money. One way to partially circumvent the educational process is to organize large organizations with the will to act as proxies for the public.

Spina Bifida in Ontario

An indication of the cost of some major chronic conditions, albeit in an indirect and not very satisfactory way is shown at Fig. 2. Very little work has gone into detailing costs because relevant data are difficult to find. They are difficult to find because there has been no demand for them and there has been no demand because epidemiologic application in the area of evaluation is in its infancy in Canada. If such costs were quantified on anything approaching a comprehensive basis, the totals would be staggering. For example, I have combined some data derived from the British Columbia Health Surveillance Registry reported by Sadovnick and Baird in 1981 with some life table estimations based on the number of spina bifida livebirths in Ontario from 1973-85 (Figs. 3, 4). A total of 1607 cases were reported in that interval and I have estimated that they lived a total of about 9,613 person-years in that time. The B.C. data show that a case of spina bifida 'costs' about $44,646 in direct medical, educational and residential terms to age 13 (1980 dollars). A cohort-specific calculation shows that these children 'cost' approximately $72 million in the time they were alive to that point. This is much less than the cost over the lifetime of this cohort and it does not include all the persons with spina bifida who were born before 1973 and were still alive. Furthermore, none of these calculations include indirect financial costs and cannot of course account for the emotional cost to the affected individual and family. This is, therefore, the incomplete financial impact of an incompletely followed, limited cohort with a single disabling condition in a single province.

An interesting additional bit of information which can be derived from Figures 3 and 4 is that survival with spina bifida seems to be improving, at least in the critical first year after birth and, contrary to popular opinion, the birth prevalence is declining very little, if at all at the present time. Constant birth prevalence with increased survival translates directly into increased costs.

Summary

Because of the high cost and serious nature of childhood physical disability, more resources must be allocated to identifying risk factors and high risk populations. Results from such studies will suggest more effective prevention and care programs. More rigorous and scientific evaluative methods are required to follow these programs because clinical impressions and other types of soft data are no longer acceptable to the medical scientific community or to the various organizations that finance the treatment of disease. Epidemiologic methods of varying complexity and sophistication have been developed for each of these objectives but they are not being applied as a matter of policy.

There is no general appreciation that sums in excess of $50 billion per year are being spent on illness in Canada (say about $2,000/person) while no more than about $10 million per year (say about 40 cents/person) is spent on the epidemiology of the same diseases. The main point is that relatively small investments in chronic disease epidemiology will almost certainly lead to very large future savings in health-related costs and human suffering.

References

1. MacMahon, B., Pugh, T.F., 1970. Epidemiology Principles and Methods. Little, Brown and Company, Boston. p.1.

2. Health and Welfare Canada. 1987. National Health Research and Development Program Inventory, 1986-87.

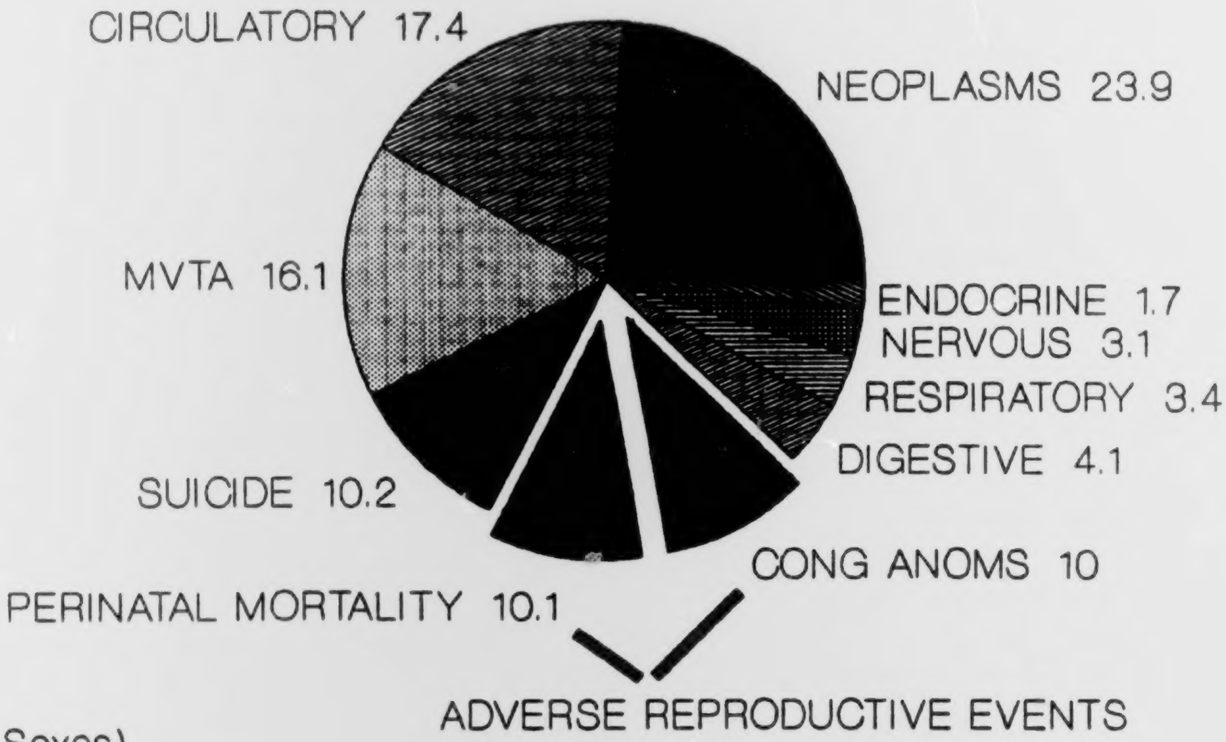
3. Sadovnick, A. D., Baird, P.A., 1981. A cost-benefit analysis of prenatal detection of Down syndrome and neural tube defects in older mothers. Am J Med Genet 10: 367-378.

CONGENITAL MALFORMATIONS
MAJOR ETIOLOGIC FACTORS

FACTOR	PERCENT
NO IDENTIFIED CAUSE	60
GENE-ENVIRONMENT INTERACTION (PRESUMED)	20
ENVIRONMENTAL FACTORS	
Maternal illness	3.5
Environmental substances	3.0
GENETIC CAUSES	
Monogenic	7.5
Chromosomal	6.0
TOTAL	100.0

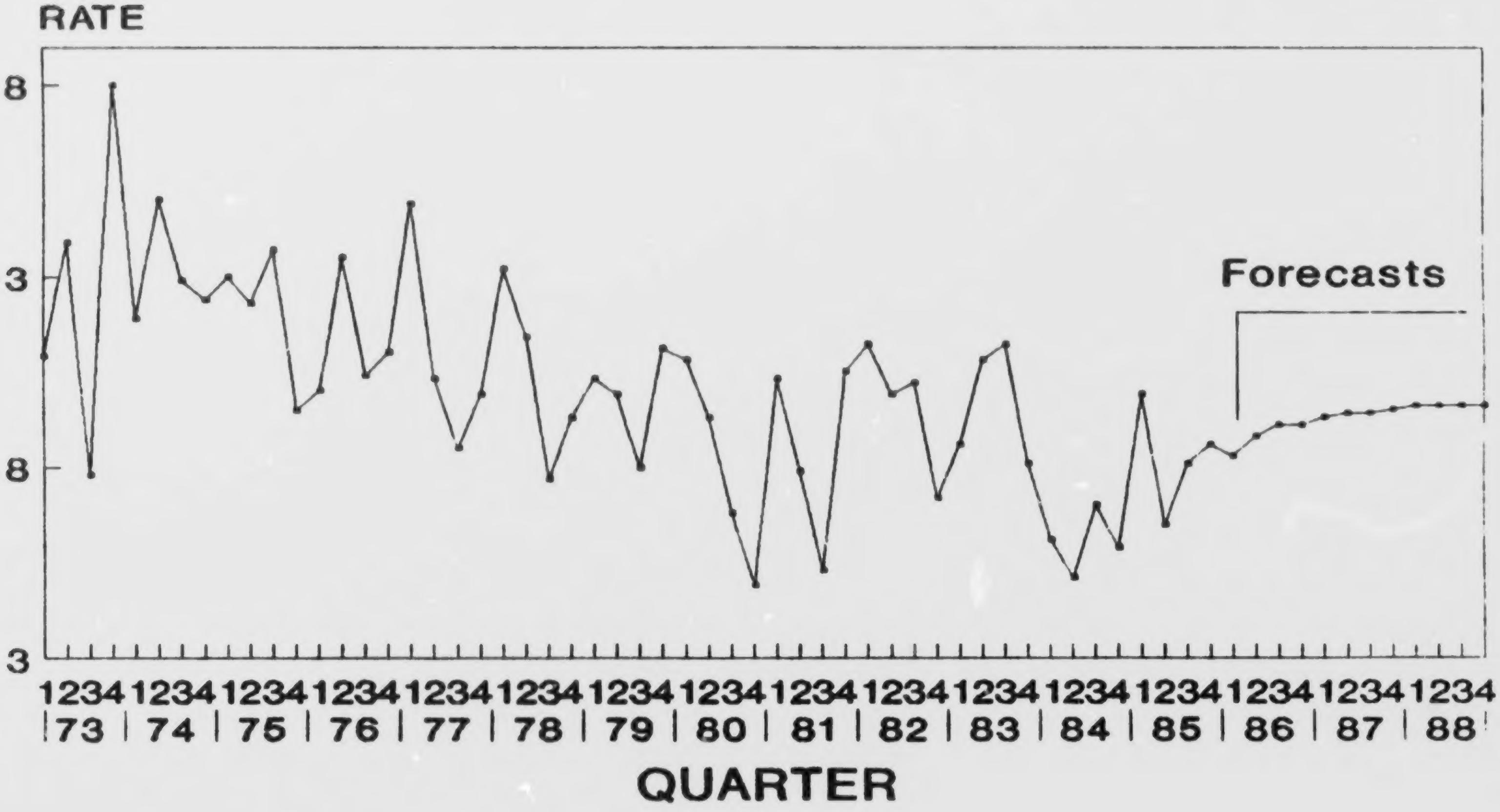
FIG. 1

TEN LEADING CAUSES OF PYLL (% OF TOTAL), CANADA, 1985



(Both Sexes)

FIG. 2



ONTARIO QUARTERLY SPINA BIFIDA RATES /10,000 LIVEBIRTHS, 1973 - 1985

FIG. 3

DEATH (%) TO FIRST BIRTHDAY IN SPINA BIFIDA CASES, ONTARIO, 1973 - 1985

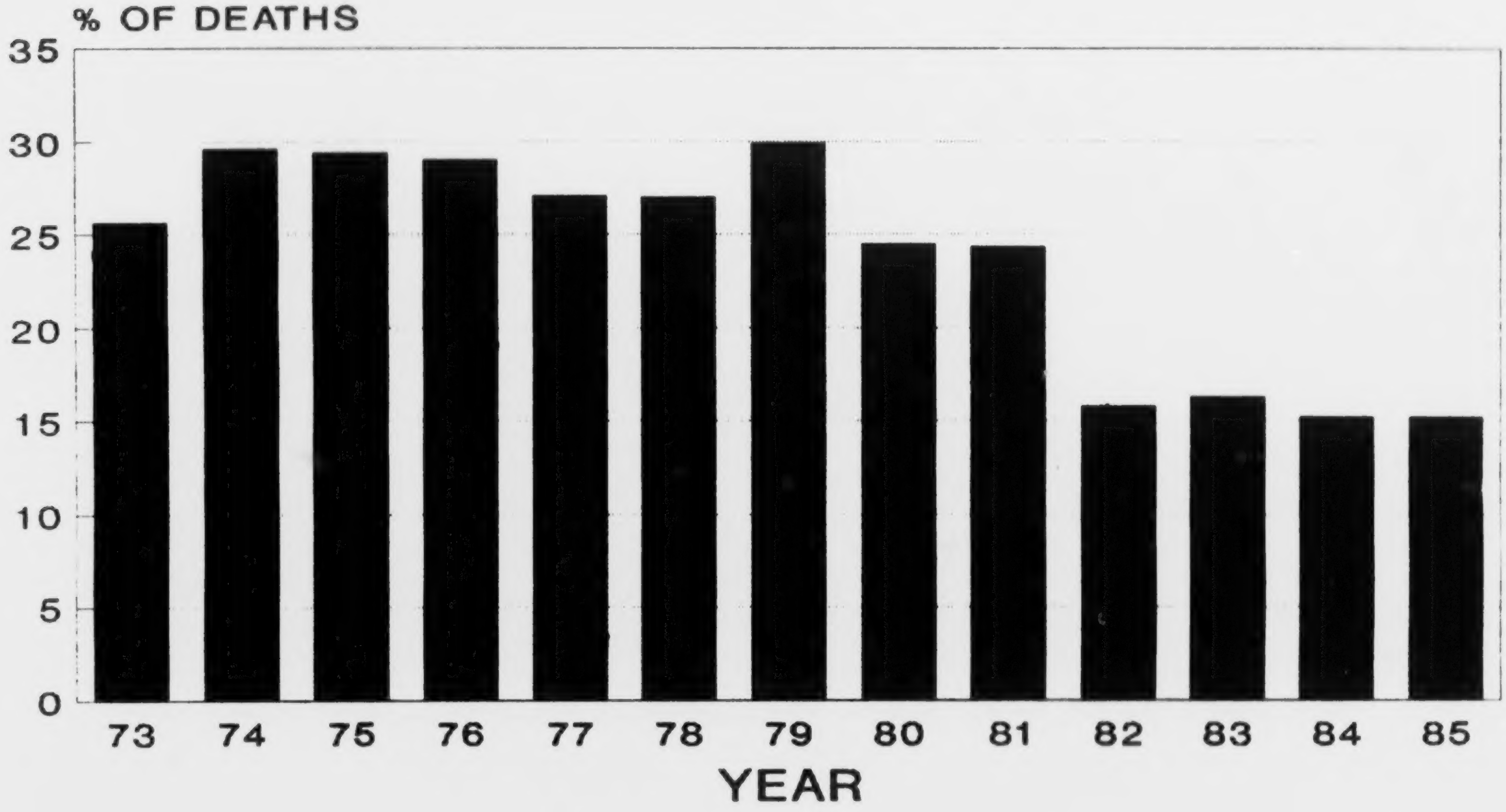


FIG. 4

5. Prevention - The molecular level

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There are a large number of research strategies and problems which fall into the broad category of molecular mechanisms of neurological development, normal and in response to all types of insults and injuries. Not all of these can be evaluated in a limited presentation. However, the discussion can be narrowed in light of the goal of identifying areas of research priority. This assignment is somewhat troubling since it would not only require an exhaustive knowledge of the current literature and extraordinary scientific judgement, but also presupposes that present knowledge provides an adequate basis to predict which directions would be the most fruitful. The history of science does not entirely argue for the success of such attempts. It does seem that the addition of time as another criterion can improve the value of such predictions when there are practical goals. In our case clinical application is the goal. Therefore, priority devolves especially to those problems and areas of investigation which are most promising by reference to earlier clinical application. It can be hoped that in surveying and evaluating the many basic science frontiers, reference to the realities of clinical problems is a reasonable yardstick by which to compare the promise of work of apparently equal basic scientific interest. I am extremely hesitant to identify any area of valid scientific advance as lacking promise for eventual clinical application. On the other hand, when speaking of the next 10-20 years, it is reasonable to emphasize certain areas on the basis of their applicability to more frequently occurring clinical problems on the one hand, of their applicability to problems whose presence can actually be recognized in the course of the human life cycle, or in the third case by virtue of their occurring at times or in ways which are more susceptible or more available to existing or putative future therapy. Guided by these criteria then, the following discussion will raise a series of molecular biological and cell biological issues and processes, each of which appears to be an important area of research priority. Their relative priority is only mentioned where particular aspects are distinguished on the basis of one or more of the three clinical criteria just mentioned.

The framework in which these research themes are presenting is the sequence of normal development. The principal unifying construct by which this discussion can currently be guided is that of unending cycle of cause and effect interactions between genetic programming and the consequences of influences impinging on all portions of the cell, from genome to plasma membrane. Most modern thinking recognizes approximately equal contributions of genetic and epigenetic or environmental influences. The contribution of the some 30,000 to 50,000 genes concerned specifically with the brain of humans offers an astronomically large basis for the seemingly endless variability of human behaviour. That this produces a base of considerable consistency in the underlying neural structures and developmental patterns does not diminish the role of a similarly diverse spectrum of interactions at all levels from the genomic to societal. The sequence of normal development is the resultant of all of these events. Like the extensive body of basic and clinical research with its modern emphasis on the richness of plasticity, this theoretical formulation supports a posture of considerable optimism for preventing or diminishing the clinical consequences of events which currently result in physical disabilities and other neurological dysfunction.

The sequence of normal development involves the essential characteristic of spatiotemporal patterning, which continues to typify the nervous system at all stages of the life cycle with regard to every aspect of its integrity. Such patterning occurs at the level of initial inductive events and in the control of the mitotic production of the nerve cells and glia. It is a feature of the developmentally regulated expression of individual genes, and of the formation and modification of neural structure and function. In the clinical context time and place are central to the analysis and characterization of both normal and abnormal function. So as not to repeat spatiotemporal issues in each section below, the general pattern in the form of a simple scheme will assist the discussion which follows. Table 1 shows such a scheme in which the overlapping and interdependent processes of normal development are conceptualized into a series of artificially separated, sequential stages. These stages will generally describe the actual sequence occurring at any particular location although both its timing in the animal's life cycle and its duration will vary markedly. There are a group of general principles applying to both development and plasticity which operate quite similarly at a phenomenological level whether one considers neurons as whole cells, dendrites, axons or synapses. At each of these levels, spatiotemporal, quantitative control is evident. The effects of this are predictable, especially in terms of outcome, provided that life circumstances have fallen anywhere within the broad range which can be defined as "normal". "Normal" in this sense does not imply that a narrowly limited set of characteristics has been present at every stage, but only that circumstances did not combine so as to exceed the plasticity available to achieve the expected outcome in the end. In other words, the reactivity induced by variations in important factors has initiated generally conservative responses, and that the capacity to respond was sufficient to dampen the variations. An example should help: how can the expected number of neurons which are normally present in a particular neural group be achieved in spite of variations in the course of development. As shown in Table 2, the number of neurons initially formed, which normally seems to be tightly controlled, can be decreased if there is a decrease in the precursor pool. The number in the final location can also be decreased depending on the success of normal migration from the ventricular zone where mitoses occur to the definitive location of these cells. Finally, the loss of neurons due to cell death, which is a widespread although apparently not universal feature of many parts of the nervous system, responds to both increases and decreases in the size of the target area of these neurons and may be diminished in the case of a decrease in the precursor pool, potentially sufficient to compensate for the decreased mitosis. Thus normal or near-normal final numbers of neurons may be reached by various combinations of such factors. It is always possible that the capacity of such responses will be exceeded. In the example of motor neurons, the homozygous state for the gene of Wernig-Hoffmann disease, or infantile spinal muscular atrophy, will lead to early and continuing loss of motor neurons beyond the amount and/or time period of available plasticity. Viewed in this way, we can also see however knowledge of the molecular events controlling the cell loss, its amount and the time period during which plasticity can continue may lead to future therapies.

The robustness of neural development is striking at the cellular level and in functional terms, including the highest functions of the cerebral cortex. However, in keeping with the above comments, robustness should be understood to imply predictable outcome, not identity at every spatiotemporal point along the way. This view seems critical if we are to anticipate how various interventions will achieve improved functional outcome even though they are applied in the setting of major aberrations or insults to the developing nervous system. It was formerly common, and remains frequent at least with regard to lower organisms and lower centers in mammals, to think of "hard-wiring" in the nervous system. This term implies analogy to a telephone network where specific wires are run from point to point, guaranteeing connections between those points while preventing the possibility of other connections through that wire. Hard-wiring is in fact a misnomer since what is actually being described is not point-to-point connections in neural development, but rather this robustness of outcome. The illusion of hard-wiring indicates how often the resultant structure and function is within normal limits, and actually retards understanding with regard to the enormous complexity and plasticity which have been increasingly documented in normal and lesion-modified development throughout the life cycle and the application of molecular insights to therapy.

The concept of plasticity has been mentioned several times. It can be reasonably claimed that plasticity and excitability are fundamental properties of the nervous system. We have moved from a time when plasticity referred primarily to changes in the mind, to current views in which plasticity subsumes changes in neural structures at all levels, changes in function such as learning and memory, their converse in the form of dementia and focal losses of capability, and the modifications of macromolecular and other chemical components of the nervous system which underlie these alterations. These processes have normal expression at each stage of the life cycle. Plasticity underlies much of the robustness of development and is a major factor in the effects of insults and injuries, including those which exceed the capacity of response. Both the normal and abnormal brain exhibit plasticity continually, so that there is an eventual blurring of recovery from injury with subsequent development and learning. The nature of plasticity at any point in the life cycle will be dependent upon the previous history of the individual and the stage of life already reached. It is now well established that changes in both basic processes and capacity for plasticity occur in complex patterns throughout the life cycle. It is certainly a long-term research goal in adult neurology to find ways to restore the capacity for plasticity of various sorts in order to ameliorate late occurring diseases, a goal which will be applicable well into childhood as well. There is of course a difference in the effects of an insult that occurs earlier rather than later. That is the cascade of effects on all subsequent stages of development, including the plasticity available for on-going modification, the cellular and synaptic effects secondary to modified tissue interactions and the circular interconnection between structure and function. To exemplify the role of life cycle on plasticity, the simplest case, let us consider the simple case of the motor neuron. The loss of an equal number of motor neurons innervating a particular muscle would have very different consequences at different times in development due to changes in plasticity. Thus the consequence of such loss would be least if it occurred before and somewhat more if immediately after migration and aggregation. It would be less if it occurred during the time of axon elongation than after innervation of muscle and the occurrence of initial cell death. It would be greater if after that, but prior to the development of skilled use of that muscle, even more so if at a time when both structure and function in the system have become stable later in childhood or in adulthood. While this type of statement is superficially compelling, one is tempted to say obvious in light of modern data, it attempts to summarize and fails to specify an enormous range of quite specific and complex neural processes including those which follow as areas of research priority.

Plasticity in the developing nervous system is known in greatest detail, and arguably is most vigorous and functionally important, with regard to synaptogenesis. Synapses have a number of quite predictable features in a given system, including where to come from, what part of the postsynaptic cell surface they contact, their neurochemical and neurotransmitter character, the size of the synaptic field of afferent neurons, and the effect of source and of target modification on these. For example, if half a retina projects to an anamniote optic tectum the axons make their usual connects, but each forms almost twice as many synapses. Conversely, a retina projecting to a half tectum forms half as many synapses. Two retinas projecting to the same tectum form their endings in alternating columns across the tectum, as do the endings of the two eyes of mammals in layer 4 of the visual cortex. These and many similar observations show a level of specificity of connections, an appearance of "competition" which probably reflects stabilization of connections related to activity, and considerable capacity for lesion induced alterations which can be advantageous, neutral, deleterious, or often a compromise with a trade off of advantage and loss. For example, crowding the termination of a full retina onto a half tectum avoids a hemianopsia at the price of diminished spatial resolution. Transplantation of parts of the nervous system, which is discussed below is a special form of plasticity, but can already be seen to follow the same rule. This fact has made it a major tool in research on plasticity.

The research priorities

Cell-cell interactions

Cell-cell interactions occur in various forms at many stages of neural development. These interactions do not represent equal priorities based on the criteria stated above. Early cell-cell interactions include those controlling the process of induction, the migration of neurons and the aggregation of neurons. It is supposed although no specific examples have yet been identified that some deleterious failures of normal neural development, including perhaps a large fraction of those which are as yet unexplained, producing some of the unexplained mental retardation and physical disabilities involving various components of the motor and/or sensory systems. Assuming that these are caused by endogenous or exogenous chemical interactions resulting in abnormalities of this class of molecular event, it is reasonable to assume that research in this area will be important for successful prevention and possibly amelioration. Later, cell-cell interactions are important for survival of neurons, dendritic elaboration, axon pathfinding, synaptogenesis, metabolic regulation, and no doubt other poorly understand processes. These will be discussed along with the three larger processes.

Induction, which has had a long history of study at a phenomenological level, denotes the influence which one cell or tissue exerts on the differentiation of nearby cells. The experimental transplantation method has provided many examples of induction, from the dorsal lip of the blastopore to lens induction by optic vesicle, and an array of variability in details of time, place and species. Comparable results have been obtained with the recent application of molecular biological methods, avoiding the effect of surgical disruptions. However, except in simpler systems such as slime molds, candidate molecules have not been identified. The newer methods do have the potential for advances, and knowledge of the molecular bases of inductive events might lead to preventative strategies, but it is not obvious how we will ever be aware of the occurrence of aberrations of such early embryological events in human gestation. Thus this must be a lower priority item in clinical terms. There is extremely limited work on inductive patterns in normal human development, and as deducible by morphological study of early human embryoes and fetuses exhibiting aberrant neural development. This long-term work remains important.

Migration of neurons from their site of final mitosis in the ventricular zone to their settling point at a distance from it, extends much later in gestation than induction. While the final mitosis of neurons in many parts of the central nervous system is completed by the end of the embryonic period proper or about 12 weeks, neurons of the cerebral cortex are generated through mid-gestation, those from the ganglionic eminence or germinal matrix through most of gestation, while neurons are still forming in the cerebellum and dentate gyrus postnatally. This later occurrence increases the possibility of clinical application. Evidence of failure of neuronal migration is frequent in neuropathological study of patients with developmental disabilities, and can now be directly visualized in the form of heterotopias with magnetic resonance imaging. There are only indirect, tentative hints of molecular mechanisms. However, well conceived and designed research should be a priority. In the near-term these will continue to be primarily studies of simple organisms from which our present insights derive. There is also a place to test normal and pathological human tissue against molecular species detected in those studies by such methods as in situ hybridization and immunoassay.

Aggregation of neurons is the end point of migration. Although not well understood, it is presumed to involve the inhibition of movement by contact between cells with matching cell surface molecules. That it is an important step functionally is emphasized by considering how cell arrangements differ in various brain structures. These differences involve the number and types of neurons present and the complex patterns of separation or intermixing of neural types, e.g. cortex versus striatum. In many structures an initial pattern is subsequently refined to reach the adult organization. An important insight into the molecular basis of such events is provided by in vitro reaggregation experiments. Beginning with neuronal cell suspensions a remarkable recapitulation of in situ neuronal groupings can occur. Some functionally related but normally separate cell groups aggregate, while separating from less related cells. This is generally interpreted as reflecting spatiotemporal patterns in the expression of the surface macromolecules controlling these phenomena. Indirect evidence is offered by discovery of an increasing number of monoclonal antibodies whose antigens are distributed in the nervous system in functionally-related sites. Some of these have been found to be localized to the cell surface. In a few cases involving early appearing molecules of more general distribution, the antigenic molecules have been identified. The role of disturbed aggregation in human disease is not clear, but probably is involved in cases of disordered relationships within neural structures, such as cerebral cortex with abnormalities of laminae or columns.

Cell-cell interactions should perhaps be viewed as a more or less unitary research area for the present. Its concepts and approaches must be applied to human diseases as well as continuing in animal and in vitro Studies. Studies of normal human development and experiments of nature can be fruitfully pursued by the methods mentioned above. Examples of this would include the study of fetal effects of maternal chemical exposure, comparing detailed analyses of controlled animal experiments to derivative analysis of human cases. The biology of the cell surface molecules under discussion should lead directly to advances in recognizing teratogenic mechanisms, and therefore relatively early preventative application. In the longer run, this line of investigation can be expected to lead to methods of altering expression of such molecules to modify or reverse maldevelopments which are detected in the course of gestation or at the time of birth. More speculatively, one can even look toward the time when re-expression of the relevant genes can be triggered in order to restore plasticity already lost due to normal developmental changes.

Dendritic elaboration

The growth, elaboration and maintenance of dendrites involves most of the mechanisms of neural development. The form and number of dendrites vary widely among different types of neurons. Since they are in part preserved under experimentally altered circumstances, intrinsic determination of general form and molecular expression is probably considerable. However, dendrites clearly require some normal interactions, particularly with afferent axons, for their full development. Dendritic elaboration begins shortly before synaptogenesis. The early form and number differ from and change over time to that characteristic of the mature neurons of each type. These modern insights have moved the dendrite from being considered a rather passive element to recognition of its active and interactive role at all stages of the life cycle. Since the dendrite is the receptive surface for a large proportion of synapses it must somehow be able to or be induced to produce and correctly localize transmitter receptors, ion channel molecules and second messenger systems. Further these have been shown to be influenced by activity, probably most effectively by simultaneous activity of the pre- and post-synaptic cells.

Of various areas of research on this problem which are current and/or likely to be fruitful, three stand out. Control of the initiation of dendrite formation and of cytoskeletal formation are principally advancing by in vitro studies which use molecular biological or cell biological probes to analyze the normal sequence and its molecular bases. The role of interactions in determining number of dendrites and details of form have been fruitfully studied in vivo. Increasing use of in vitro methods is now beginning to reveal the relation of these to the first group of studies. Most success in analyzing the control of molecular machinery within dendrites is coming from in vivo studies, for example in laminated structures such as hippocampus. Closure in terms of the interrelationships of these three aspects can already be clearly anticipated. This is an important area of research priority.

It is known that in various diseases, for example in Down's syndrome and other mental retardation syndromes, dendrites are a site of abnormal structure, and presumably of functionally important deficits. This is most striking for the abnormalities of dendritic Spine number and shape. Even if alterations in dendrites in disease are most often secondary, molecular modifications are a promising mode of therapy in the longer run. This would require that we understand some of the basic molecular events and the molecular, cellular and functional bases of dysfunction in particular patients. Thus studies of human cases are important in developing clinical applications of this research area.

Axon elongation and arborization

Soon after the completion of migration and aggregation, most neurons send out an initial process which will become its axon. Although distances are relatively small in early development and tissue barriers relatively less developed, the pathfinding of axons from their cell body of origin to their target is a striking as well as essential phenomenon. Progress in this area has come from many lines of evidence. Although little extrinsic influence is required to initiate axon elongation, two types of factors clearly are necessary. One of these is an appropriate substrate on which to grow. The other are positive and negative factors in the environment in which growth occurs. These include trophic factors, tropic factors and cell-cell contact interactions. The mechanisms of these processes are largely unknown. Yet misinnervation in limited locations is clearly common in clinical experience. There is beginning to be evidence of unexpectedly large alterations in innervation patterns in patients with early lesions, including in areas remote from the lesion. For example, cerebral palsy results in enlargement of the myotome and dermatome distributions of spinal neurons. This obviously has major importance in terms of basic mechanisms, future amelioration and in the effort to improve existing therapeutic modalities. Therefore, because of fruitful research strategies at this time and major clinical importance, this is an area of particular priority, along with the closely related process of synaptogenesis.

Synaptogenesis

It is obvious that synaptogenesis depends on the successful completion of each of the preceding stages. The interaction between the axon and the dendrite at the synapse is a two way process, although some aspects can be usefully viewed separately. yet synaptogenesis and synaptic plasticity as it occurs in normal development and learning, as well as in recovery from injuries of all degrees is to a large extent the sine qua non of the entire sequence. Therefore, this is a particularly important area of investigation. Concepts and methods are well advanced, and methods are available and advancing steadily. One of the striking insights of modern research has been the great length of time, as a fraction of the life cycle, which passes prior to the relatively stable adult connectivity is achieved in many parts of the nervous system, brain stem as well as neocortices. Even in humans, much of the normal plasticity of synapses occurs postnatally, apparently including an important role for synapses elimination as well as remodeling. There is evidence for synaptic plasticity in response to both altered structure and function in humans, as more clearly delineated in experimental animals. As in the latter that is evidence for age-related effects in the direction of decreasing plasticity. The best known examples are the eventual loss of visual acuity due to strabismus, and the relatively much greater recovery from aphasia in infancy compared to adulthood. Similar evidence exists for the motor system, for example the high incidence of facial weakness with postnatal versus prenatal hemiparesis. That this age effect reflects a change in synaptic plasticity is indicated by the fact that facial weakness is very frequent with both prenatal and postnatal lesions which produce double hemiparesis. However, the nature, degree and localization of the synaptogenetic events underlying such phenomena are not yet known. Non-invasive methods are being developed to approach these questions in patients using primarily electrophysiological and psychophysical techniques. Such research should of course be tightly based on animal experiments where numbers of subjects and analytical methods provide the basis for detailed hypotheses for the human investigations. Where possible, mathematical modeling should also be used to design critical experiments.

Metabolic regulation

The many metabolic influences between neurons and other cells and among neurons are another important class of molecular events. One knows of the major effects of thyroid hormone and insulin, for example, throughout development. Local effects are typified by the elaboration of nerve growth factor by hippocampal neurons, its retrograde transport to the cell bodies of the cholinergic afferents to the hippocampus, and its regulation there of transcription of the choline acetyltransferase gene. Along with their role in migration of neurons, glial interaction with neurons is clearest in this area of metabolic regulation. The range of problems and approaches is extremely broad. It encompasses areas the control of gene expression, protein synthesis, targeting of macromolecules within cells and much more. Spatiotemporal patterns are striking and undoubtedly important. A whole new view of the brain results from examining, for example, the distribution of a developmentally regulated antigen by immunohistochemistry. The pattern changes with time. And from antigen to antigen the pattern of staining and therefore of relationships change. The better known developmentally regulated, and regulating, molecules include the cell adhesion molecules (CAM's), microtubule-associated proteins (MAP's), and members of the immunoglobulin superfamily (Thy 1).

It is possible in a few situations to reason from clinical problems to research priorities in this area. For example, the excess of birth defects related to maternal exposure to endocrine disorders, alcohol, cigarettes, anticonvulsant medications suggest that investigation of the effects of these on embryos will be fruitful. However, much of the hope for prevention of physical disabilities due to metabolic dysregulation, and discovery of therapeutic approaches to these, lies beyond current knowledge. Yet the experimental literature on plasticity strongly suggests potential and therefore research priority must be assigned.

Control of mitosis

There is great long-term but little current prospect for understanding and modifying the complex spatiotemporal, quantitatively predictable pattern of mitosis of neurons. The most fruitful current approaches to be those which are directed at systems where the process of mitosis continues or can be restarted. These include the anamniote retina, bird song centers and the olfactory neuroepithelium of mammals. However, it seems that progress from specific neural studies will be limited until more basic progress in general cell biology points to more specific hypotheses. Since interest in cancer is a potent force in the latter, priority in neuroscience Should be low for the near-term.

Investigation of potential therapeutic measures

One of the unstable dynamics in the relationship of basic to clinical research is the question of closure. It is of the nature of basic science to lead on, seemingly forever, with every answer raising new and more complex questions. This is particularly true in the direction of reductionism, but is not limited to it. However, therapies have always been found in the face of incomplete knowledge, and this will continue. We should give priority to studies which have therapeutic potential even though we know less than desirable about their molecular basis.

Transplantation is perhaps the most striking recent example of such an area of research. Transplantation is a tool, often one among several possibilities, in basic studies of molecular and cellular mechanisms in neuroscience. It is especially powerful in developmental studies due to the requirement for immature tissue as inplant. However, it is the compelling likelihood that transplantation of neural tissue will be useful therapeutically in a variety of situations that makes it a central priority in many areas of basic research. Both the particular problem and the understanding of transplantation as a therapeutic tool are simultaneously advanced by well designed studies. Since it is likely that some transplants exert their effects primarily by providing trophic factors, others by virtue of their glial content and some by restoring a neuronal population, therapeutic use of transplants may eventually be followed by the substitution of less invasive treatments developed in part as a result of such work. Neuroscientists need no encouragement to work in this area. Support should be commensurate with the potential, realizing that high quality studies of transplantation will advance other research priorities with the same effort. Special priorities are the understanding of which cellular constituents of brain regions are needed, how to obtain these in enriched form in vitro and how to preserve them for subsequent use. It is important to consider that newborns, especially premature babies may be their own donors. This would certainly avoid some practical problems, like availability of tissue and risk of infection, as well as ethical dilemmas. But it must first be shown to be safe and effective.

There are other areas where even quite incomplete knowledge raises the possibility of experimental therapy worthy of investigation. One example is the suggestive beneficial effect of monosialoganglioside on recovery from injury. Similar consideration applies to blocking of the effects of excitatory neurotransmitters, the use of vasoactive agents to block deleterious factors after injury especially when combined with conventional physical therapy, and enhancement of recovery by trophic factors. Similarly, there are possible avenues regarding peptide neuromodulators, the critical role of phosphorylation and phosphokinases in the regulation of molecular activity. Care of course is necessary to avoid harm arising from pursuing effects without knowing whether they will be advantageous or deleterious in patients with particular problems. However, with appropriate care it should be possible to proceed with promising trials in a scientific and ethical manner, and with minimal risk to the patients who will inevitably be anxious to have the opportunity to benefit from such putative treatments.

In conclusion, we must give priority to molecular neurobiology as a major avenue to advances in the prevention and remediation of physical disabilities. On the one hand, we need to know about basic molecular events, in general as they occur in all cells, and in particular as they occur in neurons and glia. we must also know as much as possible about the nature of the molecular, cellular and functional bases of dysfunction in individual patients. Without both, we have little more than a blind chance of stumbling across a useful intervention or of recognizing it if we should. Therefore, one cannot prefer basic over clinical studies, but one can strongly argue that basic research should be frequently measured by the yardstick of applicability to clinical problems, with research directions modified by such criteria and by the insights which human disease frequently provides. In this process, our patients will be examined by and analyzed in terms of the latest methods and concepts of the basic scientist. As a result of this the natural history of diseases will be clarified. The underlying mechanisms of pathogenesis will at times be clarified. And measures for the effects of therapy will be suggested. Yet, partial knowledge and serendipity have historically been predominant over detailed knowledge in the discovery of therapies. In this setting it then becomes rational and ethical to initiate scientifically sound therapeutic trials for serious conditions on the basis of fairly tenuous hypotheses derived from basic neuroscience or clinical judgement. The challenge seems to be to do what each of us does best not in terms of priorities alone so much as in an intelligently interactive way, to accomplish the best that we are collectively capable of scientifically and medically.

References

1. Bjorklund, A., Stenevi, U., (eds): Neural Grafting in the Mammalian CNS. Amsterdam, Elsevier, 1880

2. Lenn, N.J., Plasticity and responses of the immature nervous system to injury. Seminars in Perinatalogy 11: 117-131, 1987

3. Petit, T.L., Ivy., G.O., (eds): Neural plasticity: A Lifespan Approach. New York, A.R Liss Inc, 1988

4. Purves, D., Lichtman, J.W., Principles of Neural Development, Sunderland MA, Sinauer Assoc. Inc., 1985

Table 1. Stages of development

Cell division
Neuralization
Cell migration and aggregation
Elaboration of dendrites
Elaboration of axons
Synaptogenesis
(Neurochemical and functional aspects)

Table 2. Regulation of neuron number

Number formed by mitosis - may be decreased by a decrease in the precur sor pool
Number migrating to final location
Number lost by cell death - may be greater if there is loss of part of the target area and may be less if there is loss of part of the precursor pool or an increase in the target area

Various combinations of these can produce near normal final numbers of neurons.

6. Research priorities in prevention of cerebral palsy

Reduction in the incedence of physical impairment

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Cerebral palsy must be regarded as a multifactorial condition - a final common pathway in which a large number of potential causative factors may play a role. Because of my clinical and research background, I believe my task today is to discuss the role of prenatal and perinatal factors in cerebral palsy and to suggest potential research initiatives in its prevention.

Let us consider two fairly typical examples: first, the mother who planned for her pregnancy, took great care to avoid medications, ate well, took regular exercise (not too energetic and compatible with her pre-pregnancy activity), did not smoke, stopped her regular work as a secretary two months before her "due" date, progressed through an uneventful labour and birth at term, whose baby was apparently well at birth, apart perhaps from some mild delay in establishing regular respirations after birth. The infant required occasional tube feedings, was difficult to get established at the breast and at eight months, showed definite evidence of increased tone and by twelve months, had an undoubted triplegia.

At the other extreme is the 34-year old mother of three living in poor social circumstances whose husband left home just after her fourth pregnancy was diagnosed, who smokes two packets of cigarettes a day, takes aspirin for her frequent headaches, and whose pregnancy was complicated by placental abruption at 28 weeks with precipitate birth in the small community hospital. Following transport to the Regional Centre, her 750 gram infant developed severe respiratory distress complicated by bilateral pneumothoraces. Craniosonography at two days showed bilateral Grade III haemorrhage. At follow-up, the infant was found to have a mild right hemiplegia.

In both of these instances, we would be hard pressed to anticipate the late findings and to suggest the direct underlying cause. Yet much research into the aetiology of cerebral palsy in the past has attempted to find one point in its evolution at which an intervention will be effective in its prevention. More recent reports have recognized the multifactorial nature of the condition. Unfortunately however, some reports have been interpreted as indicating that detection of adverse intrapartum status and reduction of adverse neonatal events are of little if any significance and in particular, that there is no causal relationship between them and cerebral palsy. To anyone working in neonatal medicine, regularly receiving infants from peripheral hospitals or regularly attending birth to care for the newborn, such an interpretation is difficult to accept.

Prevalence data

While research in the prevention of cerebral palsy is unquestionably important, a problem which immediately confronts the clinician/researcher in Canada interested in cerebral palsy is the lack of reliable information on prevalence. Only British Columbia seems to have attempted to develop an adequate register of children with cerebral palsy and this has only occurred in recent years. I do not wish to suggest that development of a meaningful register is easy: clinical criteria and diagnostic categories need standardization as does the age of reporting, before any progress is possible. This step is just a beginning but unfortunately at present there is no mechanism established in this province which would ensure that all children who might fulfill agreed upon criteria will be detected and reported. While I believe this step is essential, it will require a carefully orchestrated program of education for physicians, hospital nurses, public health nurses and preschool teachers.

First then, in Ontario we need facts on prevalence, clinical types and their severity.

Identification of markers during the newborn period

Attempts in the past to develop registers of infants at risk of developmental disabilities did not prove cost effective. I well recall one such register with which I attempted to work years ago in the U.K. which would have included more than 35% of all births had it been accepted.

Nevertheless, I presume that nowadays we would all agree that early detection is of benefit and that if possible, it would be valuable to be able to identify newborns at risk of developmental disability, cerebral palsy in the context of today's discussion. Some biological risk factors are well recognized, for example severe intrapartum fetal acidosis, very low birthweight, neonatal meningitis and neonatal encephalopathy. However, other markers which may predict adverse outcome also need to be assessed. The recently published report to the Minister of Health of our Province of its Advisory Committee on Reproductive Care has given some guidance in this regard. The markers suggested in that document need to be piloted and if they are found of use, to be introduced as a screening tool throughout the Province.

Thus, research in regard to markers of the neonatal period which will limit the number of children to be followed in order to detect those subsequently manifesting cerebral palsy is, I believe, a second initiative which could be appropriately applied.

Three major associated factors

In my introduction, I pointed out that for the great majority of children with cerebral palsy, attribution to a single etiologic factor is not possible. While many possible risk factors can be identified, I believe we can recognize three major associations:

1. Low birthweight
2. Fetal and neonatal hypoxic acidosis
3. Presumed intrinsic defect/congenital malformations

Low birthweight

Two major subcategories of low birthweight are recognized as at risk: the very low birthweight group, that is those weighing less than 1500 grams at birth and those who are growth restricted. The prevalence of handicap in appropriately grown infants greater than 1500 grams birthweight who escape fetal and neonatal hypoxic events is nowadays so low that they can hardly be regarded as at any greater risk than infants born at term.In very low birthweight infants and especially those less than 1 kg birthweight (extreme low birthweight, ELBW) the risk of adverse outcome remains relatively high - 5 to 10 percent in some reports of infants between 1.0 and 1.5 kg and up to 40% in some reports of infants less than 1.0 kg.

Intraventricular haemorrhage

Intraventricular haemorrhage has long been recognized as a serious complication of low birthweight infants predictive of increased risk of significant long-term disability. Technological advances have helped to clarify the relative predictive value of intraventricular haemorrhage. Computed tomography first confirmed the previous impression that outcome was related to severity. Craniosonography confirmed this but also showed that while outcome was indeed related to severity, even infants with Grade IV ultrasonic changes might have a normal outcome.

Recently, it has been confirmed that echo dense lesions with subsequent cavitation are indicative of periventricular leukomalacia. This condition was described by Banker and LaRoche as long ago as 1962. The correlation of ultrasound changes with pathology had to await the newer generation of ultrasound devices. A number of follow-up studies published in the last year or so have confirmed that ultrasound changes compatible with leukomalacia are highly predictive of cerebral palsy. Intraventricular haemorrhage and especially leukomalacia appear to result from disturbances in perfusion of critical areas of the brain.

In recent years, much research has been directed toward better understanding of factors which influence cerebral blood flow and cerebral perfusion in VLBW infants. Certain avenues have appeared promising but have either not withstood scrutiny of randomized controlled trial or have remained unconfirmed. Included in these would be the use of sedation and of paralysis in efforts to minimize fluctuations in flow and/or pressure. Likewise, disturbances of coagulation including reduced clotting factors and disturbed platelet number and function in infants experiencing severe haemorrhage have been reported, but again final proof of benefit of correcting these disturbances is awaited.

The fact that cystic periventricular leukomalacia is a critical finding in those infants subsequently manifesting motor/cognitive defects indicates a need for further search for factors influencing cerebral blood flow in low birthweight newborns and for mechanisms of controlling it. In this regard, recent technology including and PET and MRI scanning will likely be helpful.

Disturbed cerebral flow with regional hypoperfusion appears to be important in many conditions in neonates which subsequently manifest as periventricular leukomalacia. Hypoperfusion with tissue hypoxia leads to the release of free oxygen radicals which in turn appear to be important in the induction of tissue damage. Therapeutic manoeuvres aimed at reducing free radical generation and scavenging excess free radicals may offer some hope and will doubtless be addressed in length in one of this afternoon's discussions.

In many low birthweight infants, air leak syndrome complicating respiratory distress can be recognized as an important factor in the aetiology of IVH/PVL. Surfactant replacement therapy, especially immediately after birth, should in theory reduce the frequency and severity of respiratory distress and its complications and thereby reduce the neurologic side-effects. Preliminary results reported to date have not confirmed this although data recently presented by Meritt from San Diego suggest that IVH may be less severe in low birthweight infants in whom surfactant is used prophylactically. Careful follow-up of neonates treated with effective forms of surfactant will be of interest but it will be important to differentiate results of its prophylactic use from its use as "rescue therapy".

An important question which needs to be addressed in regards to IVH and PVL is the mechanism whereby risk of IVH rapidly diminishes after birth. In this, I am referring to the fact that for example the risk of some grade of IVH will be at least 60% in the infant born at 26 weeks gestation, yet one week later, the risk of a new haemorrhage occurring in an infant who has otherwise progressed well, will be markedly reduced. However, for the infant born at 27 weeks gestation, the risks soon after birth are almost as great as at 26 weeks. The $64,000 question in this area is what it is that gives rise to this rapid reduction in susceptibility after birth and the mechanisms which control it. Is it maturation of cellular integrity, circulatory stability or other factors? These questions should be a promising area for those working with animal models of intraventricular haemorrhage.

Prevention of premature birth

(a) General approach

While rasearch efforts directed at improved postnatal management to minimize complications in the low birthweight infant are unquestionably important, research to prevent birth of the low birthweight infant must be equally as important. In this regard, several questions need to be answered. In general terms, what aspect or aspects of the socioeconomic milieu of an expectant mother are important in the causation of premature labour. It is well recognized of course that prematurity is much more common in those from lower socioeconomic backgrounds. How important is nutritional supplementation in the prevention of prematurity? Convincing evidence is now available that improved nutrition in pregnancy or supplementation between pregnancies will increase the birthweight at term. However, there is as yet only suggestive evidence that these measures will have an impact on prematurity rates. Convincing evidence from appropriately conducted studies has yet to be reported. This seems to be a particularly important question to answer in view of the major efforts expended in improving nutrition of the pregnant woman. It may well be that focussed programs aimed at demonstrably malnourished women will be more effective than more general and less cost effective measures.

Specific factors associated with low socioeconomic state such as emotional stress, heavy smoking, heavy lifting and lack of adequate support systems could be found equally as important as nutritional status. In the end, it may not be possible to unravel the factors of lower socioeconomic status which are associated with prematurity but controlled introduction of interventions such as maternal support groups, smoking reduction programs and nutritional supplementation in high risk groups would certainly be preferable to a "blanket" approach which could well be regarded as unduly costly. There has been some suggestive evidence of benefit from isolated reports such as special antenatal, postnatal programs for unmarried teenagers in the States. These need to be pursued.

(b) Inhibition of premature labour

Superficially, prevention of premature labour through the use of tocolytic agents would seem to be an appropriate approach to reduction of cerebral palsy from premature birth. The problem to date has been the lack of safe and effective agents and the late presentation of women in premature labour. Isoxuprine which has been so widely used in the past, was shown only to be marginally more effective than bed rest in carefully controlled randomized clinical trials. Recent work by Dr. Lye of St. Joseph's Health Centre has suggested a reason for this. Continuous infusions rapidly saturate receptor sites rendering the myometrium refractory to further tocolysis. On the other hand, pulse tocolysis may well be more effective and trials of this mode of use of tocolytic agents are needed.

Tocolysis may well be more effective if used early in the course of premature labour. Unfortunately, reliance upon maternal perception of premature pre-labour uterine activity, has not been shown to be effective. On the other hand, the work of Creasy's group suggests that the use of a tocodynamometer designed for ambulatory care in women at high risk of premature labour may be effective. Reports of detection of threatened premature labour by use of a tocodynamometer followed by use of tocolysis indicate first, that a significant number of women are able to reach term,and second that a reduction in the proportion of very early premature labours may result. More research will undoubtedly be necessary on this aspect of control of premature labour but it appears to be a promising line. Widespread use of tocodynamometry would of course be costly and it would be important to include cost effectiveness studies in any research proposal in this direction.

Improved prenatal care for fetuses at risk

Although such features as maternal height, weight, smoking habits, parity, past pregnancy losses, medical and obstetrical complications of pregnancy, and complications at the onset of labour, are used to categorize a woman as having a "high risk pregnancy", these factors of themselves appear not to constitute reliable predictors for adverse long-term outcomes. Rather, these complications of pregnancy indicate the need for special surveillance for intrauterine growth restriction, for special attention to certain groups to minimize the possibility of premature birth, and for special care during labour and at birth to prevent hypoxia where possible.

Intrauterine growth restriction is an important cause of low birthweight which may occur at term or preterm. IUGR per se in the absence of chromosome anomalies, congenital infections and maternal alcohol consumption appears not be associated with adverse outcome. However, complications commonly encountered in IUGR infants, particularly prepartum, intra- and postpartum hypoxia, uncontrolled hypoglycemia and hyperviscosity have all been associated with increased risk of cerebral palsy. Again,research in this area would be more effective if directed at prevention rather than treatment. Studies examining the number of prenatal visits have not shown any relationship to outcome.

Unfortunately, early reception of the Ontario Antenatal Record by physicians precluded anything other than field trials of acceptability. This record includes a graph of symphysis fundal height. Nowadays, I have a distinct impression that mothers with intrauterine growth restriction are detected and referred earlier than they used to be by physicians regularly recording symphysis fundus height. Analysis of the data currently being obtained in our Outreach Education Program may in some ways help to confirm this. Unfortunately, the symphysis fundus height chart has not yet been adopted on all Canadian Antenatal Records.

It is now widely accepted that in utero transport to Regional Centres is associated with better outcomes for infants at risk than is neonatal transport. This may well relate to factors other than the transport itself including the availability of intrapartum and immediate neonatal care. The impact of a Regional Outreach Educational Program in bringing about appropriate selection and referral of mothers at high risk has not been adequately addressed. The proportion of low birthweight infants transferred within regions and the potential for prior transport of their mothers before the birth would be an accurate indicator of physician co-operation with a program in a region. There is need for research in this area, especially in a country such as Canada where small community hospitals are essential and distances and climate are major potential problems. Dr. Karen Campbell from Epidemiology and I are in the final stages of validating data from the southwest Region Hospitals which have been accumulated since 1982 which we hope will confirm the impact of our Regional Outreach Program in regard to selective referral of mothers for birth.

Asphyxia: fetal hypoxic acidosis

This problem frequently affects near term and post-term infants, especially when there is intrauterine growth restriction.Its occurrence in utero prior to labour is by no means always detected but during labour, it is nowadays detected more frequently through the use of electronic fetal monitoring in fetuses known to be at risk. In some instances, it occurs towards the end of pregnancy or labour without warning and in the absence of any antecedent factors. In the past, the use of the Apgar score as synonymous with fetal hypoxic acidosis or "asphyxia" as it would more commonly be called, led to confusion regarding its significance. Moreover, recent reanalysis and interpretations based upon the analysis of old data from the National Collaborative Perinatal project which was carried out in twelve US teaching hospitals between 1959 and 1966 has to some extent increased this confusion. Fetal hypoxic acidosis has been studied most extensively by Dr. Low of Kingston. That it is an entity which can be associated with adverse long-term outcome and that it can occur both before and during labour is, I believe, beyond doubt.

Despite the fact that the Apgar score is a relatively crude marker for asphyxia, recalculation of National Collaborative Perinatal Project data published in 1981 by Nelson and Ellenberg show that the risk ratio for development of cerebral palsy for fullterm infants whose Apgar scores were 0-3 at five minutes was 23.5 times those who scored 9 or 10. These and other data from this paper which reported univariate analysis of the NCPP study supported an association between low Apgar scores which may or may not have been indicated,and subsequent development of cerebral palsy. Using the same data set but analyzing using sequential multivirate logistical regression, Nelson and Ellenberg more re-

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more recently concluded that the events of birth and the neonatal period account for only a marginally higher proportion of cerebral palsy than risk factors identified prior to birth. This conclusion has I believe been somewhat over interpreted as indicating that perinatal asphyxia plays little or no role in cerebral palsy. As someone who has worked in neonatology for almost 30 years, I must admit I felt that it should be questioned because over interpretation is in danger of suggesting that efforts expended on intrapartum fetal monitoring or on neonatal resuscitation are futile. The infants enrolled in the National Collaborative Project were necessarily from a selected group of mothers born in prominent US teaching hospitals where even at the time of the study, attention would have been given to fetal heart rate assessment and to immediate neonatal resuscitation where necessary. Furthermore, I am assured by Dr. Campbell whose research interest is the application of logistic function, that conclusions of the univariate analysis are valid and should be given preference over results of multivariate logistic analysis. As a non statistician, even I must question methodology which commenced prenatally, and sequentially through several stages of pregnancy applied multivariate analyses which would automatically select out and attribute progressively greater statistical significance to earlier predisposing factors while reducing the statistical power of perinatal events which might occur with asphyxia. It is evident from many studies including our own that low Apgar scores do not necessarily indicate fetal hypoxic acidosis nor indeed that higher Apgar scores exclude fetal acidosis. Clinicians recognize that fetal hypoxia may occur as an undisclosed sinister event prior to the onset of labour and as such may be responsible for a proportion of cerebral palsy: animal studies of induced fetal hypoxia and acidosis are on-going, are important and are highly relevant. Fetal hypoxia occurring during the course of labour can be detected and can be subjected to research as to its significance, its treatment and its prevention. Continued research effort in this area and research on management of its resultant, hypoxic ischemic encephalopathy, must be regarded as important in both preventing and minimizing cerebral palsy in some children.

Neonatal resuscitation

If we agree that intrapartum fetal acidosis is significant and that the obtunded infant at birth needs urgent attention, it is important that we research how best this be achieved in our country with its widely scattered population, difficult climate and relatively low birth rate. Data on neonatal resuscitation in Canadian hospitals which I assembled with the help of the Canadian Institute of Child Health indicated that there was considerable room for improvement, especially in hospitals without University teaching programs. While National Guide-lines for neonatal resuscitation will soon be published as a consequence of that study, their availability will certainly not guarantee adequacy of neonatal resuscitation. precisely how the guide-lines can be applied will require substantial research into the delivery of health care to the asphyxiated newborn. How best to assure neonatal resuscitation, who should do it, how we can assist those having to do it infrequently to maintain their skills, whether more practical and effective methods than those currently used can be devised, are all questions which will require research endeavours. Furthermore, in order to assess whether any change in health care delivery to the newborn will have any impact on cerebral palsy rates, epidemiologic studies will be required, especially in non- teaching hospitals.

Presumed intrinsic and congenital defects and congenital malformations

The area of presumed intrinsic defects and congenital malformations is obviously important. In the recent analysis of the National Collaborative Perinatal Project data by Nelson and Ellenberg, these possible factors comprised the largest proportion of children with cerebral palsy. Important predictors in the NCPP study were maternal mental retardation, maternal prenatal seizures, birthweight less than 2001 grams, fetal malformations and infants presenting by the breech. Research in these antecedent factors will be difficult since they may have direct or indirect causation or maybe simply non-causative associations of cerebral palsy. I have already discussed the low birthweight association but it is of interest that in the NCPP study, of those weighing less than 2 kg, 29% had major congenital malformations. Research on brain cell growth factors and on factors which affect organization and reorganization during early brain growth may perhaps shed some light on this important group of causes. Because even with these conditions, etiology is obscure and in most instances is almost certainly multifactorial, it will be important to evaluate any effect on these conditions of any socioeconomic/nutritional interventions carried out in attempts to reduce the rate of prematurity.

Summary

In summary, I have attempted to discuss the many areas of research for which I see potential in the prevention of cerebral palsy. First, we must have accurate information on prevalence. Second, it would be valuable to try to find selected postnatal markers. Third, the multifactorial aetiology of the condition will require a multifaceted approach. Potential studies range from those of organization of health care delivery to pregnant women and their offspring, through pregnancy interventions directed at minimizing maternal fetal complications, to endeavours in the premature and full-term newborn aimed at reduction of any adverse effects of injury resulting from perinatal/neonatal cerebral insults. Additionally, studies at the cellular and molecular level will be necessary to our understanding of what are apparently important causes of cerebral palsy in selected populations.

Reduction of the incidence of physical impairment - primary prevention

Discussion

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Dr. Chance has done a masterful job of outlining the many issues involved in preventing and reducing physical disability associated with cerebral palsy. Dr. Rosenbloom will be addressing additional aspects of this problem and Dr. Banta will give us the current scene regarding neural tube defects which constitute another major fraction of the causes of congenital physical disability. Reviewing Dr. Chance's paper led me to two areas that seem worthy of further discussion. The first of these is conceptual and deals with trying to relate models of prevention to the World Health Organization's model of disease and disability. The second area is more clinical and is simply a matter of reviewing the many other types of physical disability that one encounters in children. There are many causes of physical disability in children, I will try to bring example into a brief sortie through the prevention scheme laid out for us by Dr. Chance.

Thoughtful commentators on the problems and its consequences have developed an expanded model in which different levels of prevention are defined in terms of a linear progression of the disease (Last J.M.. 1986). "Primary prevention" refers to efforts that can be used prior to the onset of a disease and may include methods such as genetic counseling, health education, or immunization. "Secondary prevention" encompasses those efforts that are directed toward identification and treatment of disease processes that are still at a subclinical or asymptomatic level. Newborn screening and treatment of phenylketonuria and hypothyroidism are examples pertinent to this conference. Primary and secondary prevention taken together relate most directly to the first segment of the WHO scheme on disability that is usually labeled "Disease or Disorder" as shown in the diagram. The juxtaposition of these two most effective phases of the prevention model with the primary phase of the disability model helps call attention to the importance of basic science research to the reduction of the incidence of physical disability in childhood.An understanding of basic biologic principles is essential to true prevention. The next phase of the prevention model, "tertiary prevention" is something of a misnomer to a clinicians way of thinking because it refers to the treatment of clinical disease. The preventive side of clinical treatment is given substance by the recognition that effective treatment begun with the clinical onset of disease may prevent the impairment and ensuing disability of unchecked illness. It follows that the clinician (and the society that employs clinicians) has a stake in making potentially curative acute care as accessible as possible to all members of society.In this regard, Canada has much of which to be proud.In the diagram I have put quotes around the term "quaternary prevention" because it is rather awkward, while the same intended notion is made much clearer by the WHO scheme. Functional disability is the main cost of illness to affected individuals and to society at large. Clinicians have often undervalued long term rehabilitative care (or in the case of children, habilitative care) for complex reasons. The WHO scheme reaches its greatest analytic strength at this stage and calls attention to the functional commonalities of all chronic disease states. In this regard, if this conference succeeds in enhancing the value of research on the prevention and treatment of functional disability in the eyes of policy makers and budget planners, our efforts here will be regarded as a watershed. Too often the rewards of dealing with long term functional disability are measurable either after the next election or in another agency's budget. This is an unfortunate double jeopardy situation. The final phase of the WHO scheme, "handicap" imposed on the disabled individual by others, has no direct counterpart in the prevention model. While I cringe at the thought of being remembered for uttering the term "quintic prevention", it must be pointed out that there is much research to be done to find ways to prevent and treat the societal and personal prejudices that handicap persons with disability. In the case of frightening conditions like acquired immune deficiency syndrome, one does not even need to be disabled before handicap impinges on the affected individual. Perhaps it would be wiser to speak of the "quintessence of prevention". In its original meaning "quintessence" refers to "the fifth or last and highest essence in ancient and medieval philosophy above fire, air, water and earth that permeates all nature and is the substance (ether) composing the heavenly bodies." For our present, less philosophical purposes, the quintessence of prevention lies in the applicability of prevention concepts to the whole gamut of abnormal phases of the appearance of physical disability in children.

I will now turn back to the outline provided by Dr. Chance's talk and discuss some specific research priorities that come to mind. In doing so, I want to call attention to the fact that there are many causes and types of physical disability in children. One can classify them from an anatomic and physiologic point of view as follows:

1. Central nervous system - brain
  a cerebral palsy
  b. primary neural tube defects(spina bifida)
  c. acquired head injury (abuse or accident)

2. Central nervous system - spinal cord
  a. spinal muscular atrophy
  b. polio & other infectious or parainfectious cause
  c. arthrogryposis multiplex congenita
  d. secondary neural tube defects
  e. trauma

3. Peripheral nerves
  a. neuropathy (genetic or acquired)
  b. trauma

4. Neuromuscular function
  a. myasthenia gravis
  b. botulism

5. Muscle
  a. inflammatory disease
  b. metabolic myopathy
  c. congenital defect (structural or ultrastructural)
  d. muscular dystrophies

6. Connective tissue and skeletal structure
  a. metabolic bone disease
  b. limb reduction disorders
  c. congenital connective tissue disorders
  d. rheumatic disorders
  e. trauma

7. Consequences of diseases in other systems
  a. nutritional disorders
  b. hemoglobinopathies
  c. etc.

Obviously there are a host of problems that still command our attention. Further headway on understanding the causes of these diseases will require us to go beyond simple notions of genetic or acquired disease to models that allow for the interaction of several factors and for social and behavioral factors.

Dr. Chance was most correct in emphasizing the importance of primary prevention of disability. Elimination of the possibility of disease is elegant, efficient and cost effective once a preventive program is established. One has only to recall the elimination of polio and the substantial abatement of rickets and tuberculosis in the developed countries of the world to understand this point. The research on which primary prevention is predicated is the most demanding. A basic understanding of diverse biologic processes followed by the synthesis of new hypotheses about these processes which can be tested using logic and objectivity is the core of the process. As we have learned yet again from the great war on cancer initiated by then President Nixon, premature implementation of applied research toward a distinct societal goal is a mixed blessing at best. The real advances may come from unexpected directions and will of necessity require the support of the apparently esoteric and irrelevant work of basic scientists, young and old, known and unknown. There will be blind alleys and fruitless efforts. Specific goals of primary prevention research include:

1. Understanding of the basic processes of fertility, implantation, placental function and onset of labour.

2. Direct and intergenerational effects of nutrition and maternal biologic and psychologic development.

3. Embryology and its deviations for a better understanding of brain development.

4. Genetics of maternal reproductive risk factors such as uterine malformation, incompetent cervix, etc.

This is by no means a comprehensive list.

Both primary and secondary prevention are well-served by research directed at the prenatal period of life. It is clear that there is much to be learned about the natural history of many disorders that begin in utero now that we are having anecdotal experience with ultrasound and other noninvasive techniques which reveal the intrauterine environment (DeVore, G.R., and Hobbins, J.C.,1979; Vintzileos et al, 1987). Basic descriptive studies have yet to be reported on normal fetal neurologic development and function. What is the role of maternal antiplatelet antibodies in the development of congenital porencephaly? What role does normal or abnormal neonatal supraspinal reflexes play in the progress of labour (Prechtl H.F.R., 1984)? We have just begun to touch this whole epoch of gestation. There are significant ethical and social issues to be understood from a research perspective if our basic understanding of biologic development is to go forward effectively.(Koop, 1986; Bayles, 1984). Perhaps the day is not too far off when we will need to define functional disability in the fetus in the same manner outlined by the WHO for adults and children.

Dr. Chance devoted space in his paper to the subject of high risk screening which is mostly applicable to secondary and tertiary prevention and to impairment and disability. I believe that the main value of high risk screening in the clinical sense is to provide feedback on how well we prevent and treat the factors that put infants at risk. Risk factor analysis also has broad epidemiologic application as Dr. von Wendt will describe in his presentation. Both of these uses of the concept of "riskness" are very valuable. Neither is a substitute for careful supervision of the health and development of every child when it is our goal to identify disability for the purpose of initiating care of individual children or studying functional disability directly. As was set forth so cogently by Dr Kenneth Holt and his colleagues years ago (Holt et al,1975; Scott, K.G., and Masi, W. 1979), it is a statistical paradox to attempt to set up a screening program for disability based on high risk criteria. If one defines risk criteria broadly enough to encompass an acceptably high proportion of the infants and children who will actually become disabled, one has an enormous and expensive task that needlessly involves large numbers of healthy infants. If one is more conservative in risk factor selection, the majority of disabled children will appear in the low risk groups even with 20-fold relative risks. Effective population screening is predicated on a clear understanding of cause and the presence of effective treatment as in the case of phenylketonuria or even congenital deafness. These issues have been discussed in detail by many people (Stangler, S.R., et al,1980; Frankenberg, W., et al,1981; Galen, R., and Gambino, S.R.,1975).

Finally, I will play devil's advocate and raise questions about some types of research that, either in purpose or method, may not be helping us move forward.I have already critiqued the notion of high risk screening of NICU graduates of specific institutions. We do need more work on the predictive value of risk factors in populations, but I would propose that we've seen too many institution-based reports already. Such activities should be part of quality-control efforts or outcome variables in experimental design studies of new NICU technologies. We have spent enough time studying abnormal pulmonary physiology while ignoring the complex socio-behavioral environment of the NICU. Have we been remiss in rushing to apply our latest technical hunch to human infants without first going back to basic reasoning and animal models?

I hope I can get away with ending this discussion with questions rather than answers. Dr. Chance has addressed Prevention as it relates to cerebral palsy in an effective systematic manner. I have attempted to abstract some crucial ideas and questions raised by his presentation that will serve as the basis of continuing discussion.

References

Last, J.M.,(ed) . Public Health and Preventive Medicine, 12th Ed, Appleton-Century-Crofts, Norwalk, Conn, 1986.

DeVore, G.R., & Hobbins, J.C., Diagnosis of structural abnormalities in the fetus. Clinics in Perinatalogy, 6:293-319, 1979.

Vintzileos, A.M., et al. Antenatal evaluation and management of ultrasonically detected fetal abnormalities. Obstetrics and Gynecology, 69:640-59,1987.

Prechtl, H.F.R.,(ed). Continuity of Neural Function from Prenatal to Postnatal Life. Clinics in Developmental Medicine-94. Spastics International Medical Pub. London, 1984.

Koop, C.E.E., Editorial. Fetal Therapy 1:1-2, 1986.

Bayles, M., Reproductive Ethics. Prentice-Hall, Englewood Cliffs, NJ, 1984.

Holt, K.S., The care of the handicapped child in the United Kingdom. In: Wallace, H.M., (ed), Health Care of Mothers and Children in National Health Services: Implications for the United States. Balinger Pub Co, Cambridge, MA, 1975:155-66.

Scott, K.G., & Masi, W., The outcome from and utility of registers of risk, In: Field, T., et al (ed), Infants Born At Risk Spectrum, NY, 1979:485-96.

Stangler, S.R., et al. Screening Growth and Development of Preschool Children. McGraw-Hill, NY, 1980.

Frankenberg, W., et al. Pediatric Developmental Diagnosis. Thieme-Stratton, NY, 1981.

Galen, R., and Gambino, S.R., Beyond Normality: The Predictive Value and Efficiency of Medical Diagnosis. Wiley & Sons, NY, 1975.

Relationship of prevention model to World Health Organization disability model

Prevention:	Primary	→	Secondary	→	Tertiary	→	"Quarternary" *		
Clinical Course	Preonset	→	Subclinical	→	Clinical	→	Chronicity		*
Who phase	*		Disorder	→	Impairment	→	Disability	→	Handicap

7. Reduction of severity

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Physical and occupational therapists have long been associated with the treatment of infants and children with motor handicaps. While therapeutic approaches have varied according to the disability being treated and have evolved with the passage of time, the overall aims of treatment are to reduce the severity of disability and to optimize function.

Many of the treatment approaches have been empirically based with little attention or concern for a theoretical foundation. This situation while not unlike other areas in medicine may be accentuated due to the nature of the population and the disabilities being treated. In most instances, children with motor handicaps are not acutely ill and are not coping with life threatening events. Rather they represent a population of individuals who are disabled or dysfunctional, not sick or ill, and who, as a group, have been devalued in our society (Caplan et al, 1987). Since there is no obvious cure or remedy for these chronic disabilities, treatments have focused upon clinical management rather than aggressive forms of intervention. Concomitant with this approach has been the unspoken belief that such treatments, while perhaps not all that effective, will at least not cause any harm. For these reasons, the demand for accountability has been less obvious than in other areas of medicine, such as drug or surgical interventions with the acutely ill.

Over the past fifteen years the call for accountability has increased. Fueled by an editorial by Taft (1972) appearing in Developmental Medicine and Child Neurology, therapists were challenged to evaluate and defend the treatment they were providing to children with motor handicaps. In this editorial Taft wrote:

> I think it is time to cry halt. I think we have an obligation to motor-handicapped infants and children, to question the entire philosophy of treatment prescriptions ... There is no hard evidence to indicate that treatment programs which attempt to modify sensory input, to inhibit primitive reflexes, or to modify or inhibit abnormal movement patterns are ever successfully incorporated into the maturing nervous system with resulting improvement in motor function. It is even doubtful if there is carry over from one day to another. Nonetheless we persist in prescribing such treatment techniques.

While this was not the first time the value of physical and occupational therapy with these children had been questioned, it might be described as the catalyst for the research efforts that have taken place over the past 15 years. A review of the literature since 1972 reveals a variety of studies that have, in one form or another, addressed the efficacy question: Does physical/occupational therapy affect the motor functioning of handicapped infants and children?

As a result of these investigations, what do we now know that we didn't in 1972? What do we still not know? What have been the shortcomings of these early attempts to answer the efficacy question? What types of research need to be conducted in the future to determine whether our therapeutic efforts are capable of reducing the severity of disability and improving function?

When reviewing the scientific literature, one is first struck with the complexity of the subject. Yet, upon reflection, it is the basic issues that, to date, have often plagued the researcher. Simply put, clinicians and researchers have failed to clarify some of the most basic tenets of treatment and therefore have been largely unsuccessful in definitely answering the efficacy question. The basic issues that in my opinion must be carefully defined in order to conduct meaningful research in the future include the following:

1. What are the specific aims of treatment? What are the expected outcomes of treatment?

2. What are the theoretical or scientific bases of treatment?

3. What are the independent variables, other than treatment, that may influence the outcomes of treatment?

4. What are the measures or indicators of the outcomes of treatment?

5. What type of research design should be used in future efficacy studies?

Aims/outcomes of treatment

While most clinicians would agree that the overall aim of treatment of motor-handicapped infants and children is the reduction of the severity of the disability and the improvement of function, there is less consensus on the specific aims of treatment. Bax (1987) in a recent editorial suggests that there is a fair amount of disagreement amongst physicians and therapists about what should be accomplished in the treatment of children with motor disorders. He proposes three general aims of treatment:

1. to prevent the development of deformities

2. to manage the child's movement to allow maximal function

3. to correct so far as possible the abnormal postures and movement patterns caused by the neurological dysfunction.

Yet, it has also been proposed that the more important outcomes of therapy should include the enhancement of 1) parental and child adaptation to the disability, 2) parent-child interaction, and 3) the child's degree of motivation and self-esteem (Harris, 1987). This lack of consensus on the aims and outcomes of therapeutic interventions has led to confusion in terms of research efforts to demonstrate the efficacy of treatments. Most previous studies have examined effects of treatment on the modification of the neurological manifestations of the motor disorders. It may well be that this particular aim is the least achievable and thus, may explain some of the inconclusive findings to date. Until, however, the aims of therapy are clearly defined and agreed upon it will be impossible to evaluate and document the impact of therapy on reducing the severity of disability.

Theoretical or scientific models

Working in tandem with the identification of the aims of treatment, therapists must be involved in the development of scientifically based and clinically relevant theoretical models upon which to base their treatment approaches. Basic research, both animal and human, is required in the following areas:

1. Neurobiology research in terms of recovery of function following insults to the central nervous system.

2. Neurophysiology and motor control research (including kinesiology and biomechanical properties of movement) in terms of the necessary components of normal motor development and motor functioning.

3. Exercise physiology research in terms of cardiovascular and muscle physiology, i.e. responses to exercise.

4. Neuropsychology research in terms of motor learning and the essential components for the recovery of motor processes following brain injury.

5. Biomedical engineering research in terms of developing assistive technology and adaptive equipment.

6. Psychosocial research in terms of impact of physical disability on patterns of social interaction.

Testable research hypotheses would then be derived from these models. Campbell (1984) proposes a departure from the traditional neurophysiologic theories of a stimulus-response model. Rather, she suggests that a central nervous system lesion in childhood of the type that typically produces a motor handicap results in three primary problems: abnormal movement, sensory deprivation, and disturbed patterns of social interaction. Each of these major areas of deficit is characterized by various components which when clearly defined provide researchable hypotheses. For example, the effects of therapy are related to the recognized effects of exercise, i.e. prevention of atrophy and the improvement of strength and endurance. Or, therapy can lead to improved social interaction as a result of increased endurance for physical activity. Campbell (1984) asserts that research designed to test these hypotheses will require different measurement tools than assessments of motor milestones or primitive reflexes. Previous studies have largely concentrated their efforts on the traditional assessments of motor development. Campbell suggests extending research outcomes to include patterns of muscular coordination, endurance, muscle biopsies, quality and extent of social interaction, self-esteem, and perceptual and cognitive abilities.

Independent variables

As early as 1962 Paine concluded via a retrospective, epidemiologic study that factors other than treatment appeared to influence the outcome of therapeutic intervention with children with cerebral palsy. Similar issues were raised in studies conducted by Wright and Nicholson (1973) and Scherzer et al (1976). Yet, twenty-five years later, there is a scarcity of information about the influence specific independent variables, such as severity of disability or socioeconomic status of the parents, have on treatment outcomes.

A variety of independent variables that my influence a child's response to treatment were outlined by Parette and Hourcade (1984) in a review article. Age and intelligence quotient have received the most attention to date, followed by the degree of impairment and degree of parental participation. Other factors posed as having a potentially significant impact on therapeutic intervention include emotional factors, time in treatment, frequency of treatment, intervention setting and type of intervention. In many of the studies these variable were simply suggested to account for the children's responses to treatment, with no statistical analysis of the proposed effects of these variable.

While the exact role any of these variable plays is still unclear, it is now evident that the response to treatment is not consistent across children and varies considerably, most likely as a result of various intervening variables. The challenge for care providers and researchers alike lies in identifying the critical variables and accounting for their actions.

Far too little attention, to date, has been given to these variables. Indeed, certain assumptions concerning these variables have been accepted without careful testing and evaluation. In my opinion, the testing of these assumptions should be a priority as part of evaluating the impact of treatment. Some of these assumptions include:

1. Early is better than late, or the earlier the better.
2. More treatment is better than less treatment, or if a little works then a lot will be even better.
3. While treatment may not be efficacious, it at least cannot be harmful.
4. Parents can be taught to "treat" their child and home program should be a part of all treatment plans.
5. Treatment will affect all children with similar handicapping conditions in a similar way.

Another way of looking at these assumptions is to develop appropriate research questions addressing the issues of timing, frequency, compliance and subject characteristics.

1. Is there a critical period in the developmental process at which to intervene which may or may not be in the "early" months of life?
2. What is the optimal frequency and/or intensity of treatment?
3. Can treatment in some instances do more harm than good?
4. Can parents really be taught to be "physical therapists" via home programs? If frequent treatments are required should those treatments be given by qualified therapists?
5. Does the quality of parenting/home environment have an impact on motor development?
6. Is there a select group of infants who are more likely to benefit from physical therapy than another group of infants? If so, who are they and what are the important patient variables?

As Ferry (1981) suggested it is time that we take a hard look at intervention programs and ask, "If intervention programs work, for whom do they work and how can they work better?" Does "One size fit all", or are certain programs better for certain children? Who should be enrolled? When? With what type of handicap? What type of therapy should be used? How often should it be given? Who should provide it? Should it be done at home, in a centre, or both? When should it be stopped? What are the potential hazards or contraindications?

Outcome measures

As earlier investigations have failed to clearly demonstrate the effects of treatment for disabled children, the cries for more sensitive outcome measures have heightened. As noted earlier, previous studies have relied almost exclusively on pre-existing quantitative measures of motor or overall development to evaluate treatment effects. With few exceptions, these measures were developed to assess overall development rather than specific components or characteristics of movement. The generally disappointing results of the few efficacy studies have often been explained by the absence of sensitive, valid outcome measures.

The development of such measures is contingent upon the development of a theoretical framework for treatment, the definition of the aims of treatment based on the theoretical framework, and the design of testable research hypotheses. Once these steps have been taken, those parameters most likely to be influenced by treatment may be identified. Reliable and valid measures to evaluate change in those parameters may then be developed. The overall goal is to have research instruments that are valid, reliable and responsive to detecting clinically important change in behaviour. What is it that we do? How do we think we impact on the children and families we treat?

Some of the parameters that deserve future research consideration include the following:

1. Quality of motor activity versus quantity of motor activity. Domains of gross motor performance include: 1) body alignment and patterns of movement including trunk rotation, balance between antigravity flexion and extension in supine, prone, sitting and standing positions, 2) movement characteristics, e.g. stability, weight bearing, or cocontraction during static or dynamic activities.

2. Energy consumption or energy conservation associated with movement and functional activities.

3. Independence in mobility and functional activities.

4. Adaptation to motor and physical limitations.

5. Quality and extent of social interaction, self-esteem and motivation.

Regardless of the parameters chosen, it is essential that the measures used are shown to be reliable, valid, and sensitive to detect change in those areas targeted as responding to therapy. Previous research efforts have been dependent on employing measures that were either 1) established essentially for other types of research or 2) were established for the specific study with little or no concern for the tenets of test construction. The development of responsive rehabilitation measures has recently been identified as a priority area by National Health Research Development Program (NHRDP). A Special Competition has recently taken place in order to encourage the development of appropriate measures. This is an important competition, and other funding agencies should be encouraged to support this type of research endeavour. Until appropriate outcome measures are available, research efforts to document treatment efficacy will be seriously compromised.

Research design

Following the review of the literature and an analysis of the studies conducted to date, one is struck with the overall similarity of the previous research efforts. While the individual studies vary in terms of rigour in research methodology, all past studies have been limited by their small sample sizes. This shortcoming is easily explained given the realities of clinical research with physically handicapped children. The very nature of the clinical population in terms of incidence, diagnoses, prognoses and treatment variables precludes any one facility from having sufficient numbers of children to carefully address efficacy questions. For that reason, it is time to mount a national/international collaborative study that addresses the efficacy question as it pertains to the physically handicapped infant/child. Pless (1976) in a thoughtful analysis and commentary on the effect of physical therapy for children with cerebral palsy concluded the following:

> Any comprehensive discussion of the value of physical therapy must consider each of the following questions: First, is all the available evidence sufficiently conclusive to warrant major changes in our management of these children? Clearly it is not. Each of the studies has been criticized on a number of counts: lack of controls, bias of various kinds, the assessment measures employed, analytic deficiencies, small sample size, etc. Many of these objections, particularly the latter, may only be met if research of the kind recommended by McGuire (1975) is initiated: more "sophisticated" protocols and fewer and larger studies. Definitive answers may only come from large-scale research conducted by a national agency, or a cooperative effort in which the work of individual investigators is developed in parallel with comparable studies in other institutions. Such collaborative ventures may be the only way to assure that the results will be sufficiently convincing that they will be incorporated into actual practice. The National Foundation or some similar body should assume the leadership that is needed to organize and conduct such a trial.

Unfortunately, a decade has passed since Pless' recommendation, and no agency has mounted such a collaborative effort. Although others have echoed the plea for larger trials, none to date have been initiated. While the need for such a trial is widely recognized, the current limitations in our knowledge preclude the organization of such a major effort. Until the theoretical basis of treatment and the subsequent research hypotheses and outcome measures have been developed, any attempt to mount a major collaborative study would be premature.

Summary

The need to assess the impact of treatment on reducing the severity of disability and improving function in the physically handicapped child has never been greater. Early attempts to address this complex question have created more questions than answers. Through these attempts we have realized that the aims of treatment are not always clearly identified, that a variety of variables other than treatment may influence outcomes, that we lack sensitive measures to assess outcomes, and that the limitations of small clinical trials often preclude accounting for all the important variations within populations. As well, our scientific and theoretical knowledge upon which to base treatment is rapidly expanding to include new interdisciplinary fields as diverse as neurobiology, neuropsychology, kinesiology, biomechanics, engineering, and exercise physiology.

As we discover the theoretical tenets of treatment, testable research hypotheses should be developed. Once established, the research hypotheses will focus the aims and expected outcomes of treatment. Efforts to design outcome measures that are sensitive, reliable and valid should be supported. At that point in time, collaborative investigations should be given serious consideration to ensure large enough populations to control for the many intervening, independent variables that may effect outcome.

References

Bax, M., Aims and outcomes of physiotherapy for cerebral palsy. Dev Med Child Neurol 29:689-692, 1987.

Campbell, S.K., Central nervous system dysfunction in children. In Campbell, S.K. (ed), Pediatric Neurologic Physical Therapy, New York, Churchill Livingstone, 1984.

Caplan, A.L., Callahan, D., Haas, J., Ethical and policy issues in rehabilitation medicine. Hastings Center Report, August 1987, pp 1-20.

Ferry, P., On growing new neurons: are early intervention programs effective? Pediatrics 67:38-41, 1981.

Harris, S., Early intervention for children with motor handicaps. In Guralnick, M.J., Bennett, F.C.,(eds), The Effectiveness of Early Intervention for At-Risk and Handicapped Children. Orlando, Academic Press, 1987.

McGuire, C., Cooperative research: necessity, luxury or stultifying straitjacket? J Med Educ 50:1131, 1975.

Paine, R.S., On the treatment of cerebral palsy: the outcome of 177 patients, 74 totally untreated. Pediatrics 29:605-616, 1962.

Parette, H.P., Hourcade, J.J.: A review of therapeutic intervention research on gross and fine motor progress in young children with cerebral palsy. Am J Occup Ther 38:462-467, 1984.

Pless, I.B., On doubting and certainty. Pediatrics 58:7-9, 1976.

Scherzer, A.L., Mike, V., Ilson, J., Physical therapy as a determinant of change in the cerebral palsied infant. Pediatrics 48:47-52, 1976.

Taft, L., Are we handicapping the handicapped? Dev Med Child Neurol, 14:703,1972.

Wright, T., Nicholson, J., Physiotherapy for the spastic child: an evaluation. Dev Med Child Neurol 15: 146-163, 1973.

8. Reduction of disability - communication technology

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As its title suggests, this paper will be concerned with the reduction of communication disability through the use of technology. It should be clear, however, that the application of technology is only one way of reducing communication dysfunction - one weapon in the armoury of speech pathologists. There are many other methods of combatting communication impairment in children (Enderby 1987), but these are not the subject of this paper. Any technology should be considered only as a part of an integrated communication environment for the client, not as the sole and only method which is acceptable. This paper is also a consideration of philosophical and strategic issues, rather than a prescriptive discourse on research priorities. There are times in the development of a field when prescription of research aims is needed: there are other times - and I believe that this is one for communication technology - when it is appropriate to look carefully at strategy. I believe that, in some senses, we are at a water-shed as far as the technology is concerned. "There is a tide in the affairs of men which, taken at the flood, leads on to fortune." (Shakespeare, W.)

Historical perspective

In the late 1950's, and early 1960's, relatively simple technology provided substantial gains in assisting the communication problems of the disabled. During the next ten years, despite a certain amount of opposition, technology gained a foothold in the management of communication impaired people. Various systems were used to great effect, though many were cumbersome, and primitive.

The 1970's provided a time when these systems came of age - the advances made in electronics served to provide more robust, smaller, and lighter systems. But these systems had been designed and built specially for a very small market, and were thus relatively expensive, or were produced in people's spare time with all the disadvantages that this brought. The turn of the next decade, however, produced yet another revolution, that of the home computer, and many of the devices which had been specially built were reproduced in software.

As this decade progressed, the increasing power and storage capacities of inexpensive computers enabled engineers to enhance already existing features, or add entirely new facilities at small extra cost. Thus, for example, the word and phrase storage, which had appeared in early systems were given much larger capacities, and the facility for synthetic speech output was added. Some systems were sold as software running on a standard home microcomputer, whereas others were specially designed (although these latter usually contained a computer within them). In both cases, the facilities potentially available to the communication impaired person were greater than the wildest dreams of the early pioneers of the subject.

The early and middle 1980's produced many and varied highly sophisticated communication aids, some of which reached the market place, and others were provided locally in small numbers. (most of the major advances were reported in RESNA proceedings).

Advanced techniques to assist the users were found in all aspects of the system. For example:

a) sophisticated input methods - head pointers, eye position monitoring, limited vocabulary speech recognition,

b) predictive and other techniques to reduce the number of input selections required, and

c) good quality, unlimited vocabulary speech synthesis.

These systems were hailed by many as "solving the problems of the communication impaired" - all that was necessary was to ensure that clients were provided with the appropriate system. As a greater range of devices became available, however, more data was gathered on their usage, and it became clear that the devices were not as popular as we had been initially led to believe. This was not, however, the "Luddite" response to technology which tended to occur a decade previously, but an informed, if disappointed view. Speech impaired people were found not to use the communication aids with which they had been supplied to anything like the extent that had been expected (Kraat 1985).

We thus approach the end of the decade with the question "What is wrong with our technology?". We need to ask "why is it not used?" and, much more importantly, "what can we do to improve the situation during the next decade?"

Power of computers

Some characteristics of our field are shared with many applications of computer technology and, as also occurs in these other application areas, there is a danger of trying to solve problems by simply "throwing computer power" at them.

Because of the enormous increase in computation power available within systems over the past twenty years, it is possible to extend concept much further than had hitherto been possible. In this situation, there is a danger of pursuing the development of an idea long past the point of diminishing returns - plowing a particular furrow long after the increase in returns are justified. Early communication devices, for example, were able to store a few short sentences - modern devices can and do store thousands of sentences for later retrieval by the user. It is, however, not clear that users will be able fully to exploit the enormous storage capacities of some current machines. Increases in computational power make it a great deal easier to implement even very complex ideas on a small portable computer (Some examples of current ideas which come into this category can be found in AAC 1987). A much greater range of ideas is feasible than has been the case in the past, and it is often much easier to implement ideas using modern computer-based technology. The problem of ensuring that designers do have good original ideas, however, remains.

What is wrong with our technology?

There is really only one answer to the question "Why are our devices not being used?" - it can only be because they are not seen to satisfy the real needs of customers, always remembering that "customers", include, not only the speech impaired, but also those with whom they communicate, and those who prescribe communication systems (Newell, 1987).

Many of the reasons for failure may appear to be non-technological. Although the speech impaired person may have a system which allows him to "speak messages" he experiences difficulties in using it appropriately in real conversations. This can be due to insufficient training in the use of aids, but also can be caused by inexperience in the rules of discourse. "Having the right tool does not mean that one automatically becomes a skilled craftsman" (Easton, 1987). However, we are not yet sure of how or what to teach. In addition, perhaps we do not yet have systems which are effective in a conversational environment. Have we yet designed a tool which even a skilled craftsman can use effectively and efficiently?

Yoder and Kraat (1983) reminded us that augmentative communication aids are "not just different vehicles that are comparable to speech. They are uniquely different systems with different capabilities" and we are only just beginning to take full cognisance of this. But the field is young, and the problems being tackled are very complex. It is thus hardly surprising that a definitive solution has not been found. Technologists can fly us to the moon, but their success is based on science having had a very clear understanding of the problems involved for many years. In contrast, the field of communication systems for the speech impaired is closer to a pre-Galileo phase of understanding.

The need

I believe that what is required in this field is some radical thinking that will lead to some good new ideas. However the production of new ideas cannot be turned on like a tap, nor can a single author list the new ideas which should be generated.

"I can call up spirits from the dead, but will they come when I call?" (Shakespeare, W.)

The major useful action which can be taken, however, is to try to create an infrastructure and atmosphere which encourages new ideas.

The strategy one adopts in such a situation critically depends upon the time-frame in which a solution is expected. A strategy which will create a solution for a particular child by the end of the month is radically different from one which may produce a new system within a year, which is again different to one which is appropriate for a five to ten year research project. Interaction between people working in these different time-frames is essential, but equally essential is that each worker should know what is his or her primary time-frame.

This paper concentrates on the long term needs for research in this field, and the comments are thus mainly applicable to the five to ten year time-frame.

Characteristics of an appropriate research strategy

A research strategy with a five to ten year time-frame should not be too closely focussed on today's perception of clinical needs. Unless the customer population is very enlightened, their perceived needs are often articulated as the detailed inadequacies of current systems. This usually implies day-to-day and month-to-month problems, and these are properly dealt with in a service environment. This service function is, of course, vital and should be linked to research environments to their mutual benefit. Because the needs of the two environments can be very different, however, it is counter-productive for short term needs to be allowed to dominate a research environment.

With a five-to-ten year time-frame;

i) The primary interest should be in radical change, the effects of which cannot be fully foreseen,

ii) Good ideas are unlikely to come entirely from within the boundaries of the discipline as it is currently perceived. They will break new ground and be contrary to "received" wisdom

iii) Thus, unless an idea creates at least some scepticism, or even hostility, it is unlikely to be really novel. (Early motor cars were seen as somewhere between a threat and a device of the devil, and certainly useless - in its early days they had very few supporters)

iv) It is difficult to predict losers and impossible to predict winners in these situations.

We thus need to encourage a wide perspective in our researchers clearly a thorough knowledge of current practices - but also a healthy scepticism, and we must be prepared to back their hunches. Although we need to keep our researchers' "feet on the ground", we should be wary of condemning new ideas too readily. It is too easy to dismiss radical solutions, and demotivate a good researcher.

Similarly, funding agencies should be wary of being too conservative and only supporting work which will clearly be successful - The future is a gamble and the longer the time-scale the longer the odds.

The requirements for a research team

The requirements of a research team in this field include:

i) The team should be fully familiar with but not dominated by the service needs,

ii) Particularly in view of the current trends in technology, researchers should spend a greater proportion of their time thinking about what they want to achieve rather than how they are going to achieve it.

iii) The team should be fully multi-disciplinary. The work is too important and complex to be left to engineers or computer scientists. Speech pathologists should be encouraged to take a more active role in the design process, and become fulltime members of the design team, an educationalist should also be represented on the team.

iv) Advantages would accrue from including representatives from unfamiliar disciplines: Linguists, anthropologists, philosophers, sociologists, graphic designers, and people with experience of "communication media", such as television.

Focus for research

As has been stated, the root problem is that current systems do not appear to satisfy the real needs of our clients. It is easy to say where work is needed. This obviously includes the following;

i) Improving the rate at which systems can be operated by developing;

- a) Efficient and cosmetic non-mechanical input devices,
- b) Systems which are more effective in predicting the messages which are required to be transmitted.
- c) Effective coding and/or access systems for large data bases of messages, and
- d) Non-linguistic access to message data bases.

ii) Improving the acceptability of the messages which are transmitted by;

- a) Improved large vocabulary speech synthesis,
- b) More appropriate visual display systems.

iii) Improving the systems aspects of devices by:

- a) Making systems more effective for promoting appropriate conversational behaviour (turn taking, maintaining place, feedback, initiating and encouraging conversations) rather than simply delivering messages.

b) Improving cosmetic aspects of the systems - Would I like to have one in my home, on my wheelchair, on my person?

Ascertaining the real need

Although the above suggestions may be helpful in providing some pointers for research in the future, the major research questions are:

"How do we ascertain what are the real needs?" and "How do we encourage our researchers to set goals which they might be able to achieve"?

Strategies for research teams

There are a number of strategies which could be used. The biggest danger, however, is to concentrate on only one strategy. we are all in such an appalling state of ignorance that we need to encourage a plurality of strategies. Some suggested strategies are;

i) Focus perception of needs on the imagination of the designers rather than their perception of the needs of the clients. "What are the characteristics of a communication system which you would design for yourself?"

ii) The above suggestion would be made more effective by encouraging researchers to find out personally what it is like to communicate using current alternative communication systems in a range of conversational environments.

iii) Go to other disciplines to see what they have to offer. Suggestions of such disciplines have been given earlier, and research teams have already gained advantages from detailed studies of, for example, discourse analysis.

Encourage links between groups

It is probably unrealistic to produce a research Group with representatives from all the appropriate disciplines. Also, due to the different composition of the teams, cultural differences, and their exposure to different clinical situations, different research groups will have different approaches to problems. It is thus important to promote links between groups which encourage the interchange of ideas and of researchers. Resources need to be allocated specifically to this activity as it can be a very effective way of proceeding in a relatively young interdisciplinary field like ours.

Conclusions

Few people are capable of accurately prescribing the details of an appropriate long-term research strategy, particularly when the field is in a state of flux. In this situation, we should not try to prescribe particular goals, but concentrate on offering suggestions as to the best ways of achieving goals. This paper has tried to do the latter - we need to rely on the vision of our researchers for the rest. Perhaps the best we can hope to do is to point them in the right direction, and not stand in the way of new radical ideas. It is also essential that we are there to encourage them when their ideas do not seem to be working and when even they are at a low ebb.

References

Kraat, A., 1985, "Communication Interaction between aid users and natural speakers; a state of the art report." Canadian Rehabilitation Council, Yonge Street, Toronto, Canada.

Enderby, P., Assistive Communication Aids, Churchill Livingstone 1987.

Easton,J., "Developing effective communication in aid users", in "Assistive Communication Aids" Ed. Enderby, P. Churchill Livingstone, 1987.

RESNA; Proceedings of the Rehabilitation Engineering Society of North America. 1978 - 1987.

AAC; Augmentative and Alternative Communication - Special Issue Vol.3 No.4 Nov.1987.

Newell, A.F.,"How can we develop better communication aids?", Augmentative and Alternative Communication, Vol. 3, No. 1, March 1987.

Yoder, D.E.,& Kraat, A., "Intervention issues in non speech communication; Contemporary issues in language intervention. American Speech, Language and Hearing Association Report 12.

9. Priorities for research in the field of augmentative communication

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The field of augmentative communication has experienced tremendous growth in the past two decades and has provided increased independence and improved access to communication for physically disabled children who are unable to speak and/or write. In order to ensure that clinical and educational programs are optimally effective in meeting the complex needs of this population, concerted clinical research is required to develop a sound, empirical base of knowledge in the field, while technical research and development is needed to forge a new generation of communication systems and supporting technologies. Specifically, clinical research is needed to address the following priorities: demographic research to identify the characteristics and needs of individuals who use augmentative communication systems and to determine the training and resources needed by their facilitators; research to guide in the selection, customization, and integration of augmentative communication systems; research to identify factors which determine the communicative competence of augmentative communicators; research to determine vocational opportunities and to delineate the requirements for educational curricula; and research to evaluate the efficacy and cost effectiveness of clinical, educational and vocational training programs. Future research and technical development should be determined by the needs and skill requirements of clients using augmentative communication systems. Priorities for technical research and development include the following: research into the technical and functional integration of technology to meet the communication and daily living needs of disabled individuals; research to develop communication systems which provide greater flexibility and simplicity in their design; research into innovative access methods to better accommodate the needs and skills of users; research and development to ensure that standard computer-based equipment is accessible to disabled users; research to develop more efficient acceleration techniques to enhance the rate of communication for physically disabled clients; research to improve the quality of speech output and to provide the option for multi-lingual output; and research to allow access to telecommunications.

Priorities in research in the field of augmentative communication

It has been estimated that there are approximately 200,000 nonspeaking individuals within Canada (Milner & Parnes, 1984). Many of these individuals are children and adolescents with physical disabilities which preclude functional spoken or written communication. Without access to conventional channels of communication, these individuals are severely limited in their ability to participate fully within family and community life and within educational and vocational pursuits. As a result, they are significantly at risk in their cognitive, social and emotional development, for it is through communication that individuals acquire knowledge, initiate and sustain interpersonal bonds, and express their needs and wants.

Over the past two decades, important developments in the field of augmentative communication have provided many nonspeaking physically disabled children with more effective means to communicate, including unaided systems such as gestures and sign language, and aided systems such as communication boards of pictures, symbols, alphabet and/or words. Recent technological advances have led to a range of computer-based communication devices developed to meet the face to face and written communication needs of nonspeaking persons. Despite the potential benefits offered by these augmentative communication systems, clinical observations and research findings indicate that most nonspeaking physically disabled individuals fail to make full and effective use of either nonelectronic or electronic communication aids in their daily interactions, (Kraat, 1984).

Historically, the field of augmentative communication has been largely clinically based. The growth of the field has been evidenced in the inception of service delivery programs and in the development of augmentative communication technologies. While these advances have been important ones, at present, clinical practice is largely driven by the intuition and experience of the transdisciplinary assessment-intervention team. To date, there has been a paucity of research in the field to guide clinical practice. Mathy-Laikko and Yoder (1986) have noted that by far the majority of the documentation in the field of augmentative communication reflects review articles and descriptive reports of the "possible"; less than a third of the articles are research reports. Moreover, the focus of the latter reports has been a narrow one. There is a critical need for concerted research in the field of augmentative communication to develop a sound empirical and theoretical base of knowledge to guide clinical practice and to ensure that assessment-intervention approaches are effective and cost efficient. The intent of this paper is to suggest priorities for future research in the field from both clinical and technical perspectives.

Priorities for clinical research

Prutting and Kirchner (1987) have argued that "our effectiveness as clinicians is judged...by the impact our remedial efforts have on an individual's ability to function as a productive member of society."(p.115). Clinicians and educators in the field of augmentative communication face a significant challenge in providing the augmentative communication systems and skills training required to ensure that nonspeaking physically disabled children develop the communicative competence and independence required to realize their full potential. In order to effectively meet this challenge, clinicians and educators require access to a sound information base of empirical research to support their decision-making role in the assessment of clients, the prescription of communication systems, the development of appropriate intervention programs and educational curricula, and the evaluation of the efficacy of these programs. Given the recent history of the field of augmentative communication, it is not surprising that the empirical information base is scant. It is only within the last few years that there has been any systematic research activity. Much of the available clinical research has consisted of descriptive studies. While this research has contributed to a better understanding of some of the issues in the field, it has been plagued by methodological problems including the problems of a heterogeneous population and small 'n' studies, the lack of valid and reliable measures, the absence of a theoretical base, and the intrusion of confounding variables. Concerted research activity is required to expand the currently available information base and to guide the decision making of clinicians and educators. Specifically, research is required in the following areas:

1. demographic research to identify the characteristics and the needs of individuals who require augmentative communication systems;

2. research to guide in the selection, customization, and integration of augmentative communication systems;

3. research to identify factors related to the development of communicative competence by augmentative communicators;

4. research to delineate the requirements for educational curicula and to determine vocational opportunities for physically disabled individuals who use augmentative communication systems; and,

5. research to evaluate the efficacy and cost effectiveness of specific intervention programs and of various models of service delivery.

Each of these five areas is discussed in more detail below.

Demographics of the population

To date, there have been only a few studies which provide any demographic information concerning the population of individuals who use augmentative communication systems. Matas, Mathy-Laikko, Beukelman and Legresley (1985) published a demographic study of the nonspeaking population in the state of Washington. U.S.A., while Aiello (1980) and Lindsay, Cambria and McNaughton (1986) reported on unpublished demographic studies of school-age nonspeaking students in Orange County, California, U.S.A. and in the province of Ontario, Canada, respectively. Although these studies have made initial contributions to documenting the characteristics of the population, further demographic research is required to consider wider geographic areas and to provide more detailed information in the following three areas:

1. the characteristics of the population and the available services;

2. the needs of the population; and

3. the training and information required by their facilitators, that is, the significant others in the client's life, including parents, siblings, teacher, teacher's aide, residential counsellor and/or members of the community-based (re)habilitation team.

Characteristics of the population

Research is required to document the following characteristics of the population of individuals who use augmentative communication systems: age, diagnoses, concomitant impairments, geographic distribution, place of residence, family characteristics, educational or vocational placement, available support services, augmentative communication systems used, skill levels achieved, etc. This information is essential as a basis for justifying the need for funding to support clinical service and research activities focused on the needs of this population. The availability of demographic information will greatly facilitate service delivery planning by identifying the geographic distribution of the population, the available services, and client needs and characteristics. Demographic research will also serve to identify neglected segments of the nonspeaking population (e.g., multi-handicapped individuals, head injured individuals, and adults) and to ensure that service delivery programs and research programs address the needs of these groups. Research is also required to identify client and/or familial factors which place children significantly "at risk" for speech development in order to support early identification and intervention with infants who may require augmentative communication systems and with their families.

Needs of the population

In order to be optimally effective any service delivery must be driven by the needs of the clients and families served. It is only by fully delineating the client's communication needs that it is possible to provide a clear focus for intervention to maximize the client's functioning in the environment. As Beukelman, Yorkston and Lossing (1984) have noted, neglecting to account for the client's needs may result in intervention that fails to produce a functional impact. While it may seem to be a relatively trivial matter to determine the communication needs of individuals who require augmentative communication systems, in fact, the process is not nearly as straightforward as it first appears. Communication needs may vary depending on the demands of educational/vocational, residential, and recreational environments, on the partners involved, and on the personality and life style choices of the individual client.

To date, there has been minimal research to document the communication needs of the population. There are few assessment tools to assist the clinician in determining the communication needs of clients. Available instruments tended to be limited in the scope of their application (e.g., Dowden, Honsinger and Beukelman, 1986, have developed a communication needs assessment for use with patients in acute care settings). Further research is required to articulate the range of communication needs for other groups of individuals who use augmentative communication systems including children and adolescents, individuals who are ambulatory and those who are not, individuals who are integrated into the community and those who reside in institutional settings, etc.

This research should document the broad interaction or social needs of the client, as well as specific message, contextual, positioning, partner, system, and modality requirements. Needs assessments which deal only with specific message needs at the surface level of the interaction run the risk of determining intervention which facilitates the communication of specific, routinized messages, but which misses the client's broader social agenda. As Light (in press) has noted, to date it has largely been assumed that communication to express needs and wants and to exchange information is of greatest importance to physically disabled individuals who use augmentative communication systems. Little attention has been directed towards the need for social closeness and towards intervention to facilitate such communication. If the need to promote social closeness is not taken into account, intervention and system prescription designed to bring greater efficiency and effectiveness to information sharing and to the expression of needs and wants may at times result in impediments to the goal of social closeness.

Research is required to develop protocols for assessing the communication needs of individuals who use augmentative communication systems. The information gleaned through these needs assessments can be used on an individual basis to guide clinical intervention and system prescription. On a more global level, this information can be used to provide functional specifications for technical developments in the field, thus ensuring that the future generation of augmentative communication technologies meets the needs of the target population as effectively as possible.

Training and information required by facilitators

Successful communication depends not only on the skills and systems available to the augmentative communicator, but also on the opportunities for communication and the support provided by facilitators, that is, the significant others in the client's life. In order to provide appropriate opportunities and support, facilitators require access to information regarding augmentative communication systems and access to training in interactional strategies and techniques. In order to develop service delivery models to effectively deal with the needs for facilitator training, demographic research is required to determine the characteristics of the facilitators of augmentative communicators, their experience and training, their current levels of expertise, the support networks and resources available, and their needs for further support, training, and resources. This information would serve as the basis for improving currently existing parent/facilitator training programs and professional workshops. It would also provide the impetus for developing augmentative communication training programs within the universities for undergraduate, graduate, and post-graduate level education of professionals, as well as for developing continuing education programs to upgrade the skills of practicing clinicians and educators.

Selection, customization, and integration of augmentative communication systems

Learning to communicate effectively by means of augmentative communication systems can be compared to learning to play a musical instrument. In both cases, there are two essential components: the tools (e.g. the augmentative communication systems or the musical instrument); and the knowledge, judgement, and skills required to effectively use these tools. These components are mutually dependent: one without the other is insufficient to ensure a competent performance. Research is required into both of these components: research into the selection, customization, and integration of augmentative communication systems to meet the client's needs given his/her skills; and research into the knowledge, judgement, and skills which determine communication competence in daily interactions. This section of the paper addresses the system-related research issues, while the subsequent section of this paper deals with the research issues related to communicative competence.

As Mathy-Laikko and Yoder (1986) have argued, to date, "we have largely ignored the impact of communication aids, special symbols, strategies, and techniques on the overall process of communication." (p. 479). Yet it is essential that studies be directed towards determining the strengths and constraints of augmentative systems. This research needs to address two central questions:

1. What are the user requirements imposed by various augmentative communication systems?
2. What is the impact of these systems on the client, the partner, and society at large?

The user requirements imposed by augmentative communication systems

Use of any augmentative communication system, be it a sign language or gestural system, a communication display of Blissymbols, or a computer-based voice output communication aid, imposes specific motoric, cognitive, linguistic, and sensory/perceptual demands on the user. Much of the early research work in the field of augmentative communication considered the demands posed by some of the available augmentative communication systems. (See Doherty, 1985, for a review of the literature on the effects of sign characteristics on sign acquisition and retention and see Vanderheiden and Lloyd, 1986, for a review of the literature on the characteristics of aided and unaided systems which affect learning). These research incentives, although important ones, have touched but a few of the many issues posed in the selection and integration of augmentative communication systems to match client skills and needs. The research efforts in the field have by no means kept pace with the ever-increasing technical developments which have resulted in a proliferation of options, including various symbols, vocabulary organizations, selection techniques, processing capabilities, and output modes. As a result, there is only a limited understanding of the human factor issues involved in the use of augmentative communication technologies.

While technological developments offer the potential to by-pass many physical limitations and to increase independence and function for individuals who require augmentative communication systems, this potential will only be realized if clinicians have full understanding of the motoric, cognitive, linguistic, and sensory/perceptual requirements for system use and are thus able to optimize the match between the client's skills and needs, and available technologies. Unfortunately, the literature indicates that augmentative communication systems tend to be selected on the basis of clinician familiarity and system availability, rather than according to a systematic assessment of client and environmental needs (Fristoe & Lloyd, 1978). Too often, clinicians resort to "trying on" available systems with the client in a haphazard attempt to determine the "best fit". With the rapid technical developments in the field and the proliferation of available systems, this trial and error approach to system prescription is no longer reliable, effective, or clinically viable (Microcomputer Applications Programme, 1988). Thus, research is required to determine the user requirements imposed by augmentative communications systems, to develop protocols and expert systems to support clinicians in the process of system prescription, and to evaluate instructional strategies to teach clients to use the augmentative communication systems prescribed effectively, efficiently, and accurately.

The impact of augmentative communication systems

In the prescription of augmentative communication systems, it is not sufficient to consider only the skills required by the user, rather clinicians must also consider the impact of the systems on the client, partners, and society in general. In the past, it was assumed that the augmentative communication system would simply replace the individual's deficient speech function. It is now generally recognized that this assumption was entirely erroneous: use of augmentative communication systems has fundamental implications for rate of communication, independence, content of communication, perceptions of the partner, etc., which render the resulting interaction unique from models of spoken interaction (Yoder & Kraat, 1983). Research is required to determine what effect manipulations of augmentative communication components (e.g. output mode, vocabulary, selection techniques, etc.) have on the Communication process (Mathy-Laikko & Yoder, 1986). Some initial research has been conducted in this area: for example, research has considered the attitudes of listeners towards various output modes (Coxson & Mathy-Laikko, 1983), the intelligibility of synthesized speech and listener preferences (Mirenda & Beukelman, 1986), and the preferences of partners for interactions involving nonelectronic versus electronic communication systems (Buzolich, 1984).

Research to delineate the impact of various augmentative communication system components should consider all three perspectives within the interaction: the client perspective, that of the partner, and that of the third party observer. As individuals who use augmentative communication systems are increasingly integrated into educational, vocational, and community settings, research is required to determine social attitudes and perceptions towards clients using various communication systems. Through a better understanding of factors which influence social perceptions, it will be possible to determine strategies to minimize the "handicap" imposed by society on individuals who use augmentative communication systems (Warrick, 1988).

The research to date has indicated that communication is largely a multi-modal process for individuals who use augmentative communication systems (e.g. Kraat, 1985; Light, Collier & Parnes, 1985). The modes used seem to vary according to the context, the partner, the message (its content and intent), and the social dynamics of the interaction. Through research to determine the benefits and constraints of various augmentative communication systems, it will be possible to ensure that the systems prescribed for a client are truly meeting the contextual, partner, message, and social requirements and that they are thus contributing to greater communicative effectiveness for the client.

Factors related to communicative competence

Historically, intervention in the field of augmentative communication focused on teaching the client the mechanics of system use, often outside the functional context of daily interactions. Despite these interventions, clinicians and researchers observed that augmentative communication systems tended to be poorly utilized and underutilized by clients to meet their communication needs within the natural environment (Kraat, 1984). It has now been recognized that proficiency in the operation of an augmentative communication system does not, in and of itself, ensure the development of communicative competence by the user (Kraat, 1984). Rather, concerted intervention is required to ensure that the client develops the necessary knowledge, judgement and skills to become an effective communicator.

Most currently available assessment instruments and intervention programs tend to be premised on normal models of spoken communication. These approaches have proved to be counter productive, since they emphasize the client's deficit without clearly indicating appropriate channels for functional remediation. As a result, several researchers and clinicians in the field have argued for the need to develop functional models of communicative competence which account for the differences inherent in the interactions of individuals using augmentative communication systems (e.g. Buzolich & Higginbotham, 1985; Kraat, 1985; Light, in press). To date, however, there has been little research to articulate a paradigm of communicative competence for individuals using augmentative communication systems and to test out the theoretical model to ensure its validity in the field.

Recently, Light (1988a) has argued that the development of communicative competence by individuals using augmentative communication systems is predicated on the development of knowledge, judgement and skill in four interrelated areas: linguistic competence (i.e. adequate mastery of the linguistic code of the native language(s) spoken by the community and of the "linguistic" code of the augmentative system); operational competence (i.e. accuracy and speed in the use of the augmentative system); social competence (i.e. sociolinguistic and sociorelational competence); and strategic competence (i.e. adaptive strategies to overcome linguistic, operational, and social limitations). The former two factors (linguistic and operational competence) reflect knowledge and skills in tool use, while the latter two factors (social and strategic competence) reflect functional knowledge and judgement in interaction. The definition of communicative competence proposed by Light suggests at least four factors which may be related to the development of communicative competence by individuals using augmentative communication systems. Research is required to investigate the validity of this model and to identify other factors which determine the communicative competence of clients across various partners (familiar and unfamiliar, adult and children), within various social contexts (formal and informal, dyadic and group), and across various environments (home, school/work, community). Research is needed to develop valid and psychometrically sound measures to assist the transdisciplinary team in assessing the communicative competence of individuals using augmentative communication systems and in developing effective intervention programs. Finally, research is required to develop effective training paradigms for teaching specific skills and to evaluate the effect of this skills training on the communicative competence of users.

Just as research is required to identify factors which contribute to the communicative competence of individuals using augmentative communication systems, so too is research required to identify the skills and strategies which contribute to the effectiveness of facilitators as partners in the interaction. The latter research will provide the empirical base for developing the content of facilitator training programs to optimize the facilitator's skills in supporting the client's daily interactions. This future research should be directed towards identifying factors which determine the communicative effectiveness of augmentative communicators and of their facilitators. This research will ensure that clinical assessment-intervention is optimally effective by identifying the issues of critical importance for clients and facilitators in their development of communicative competence.

Educational curricula and vocational opportunities

Too often in the past, physically disabled individuals requiring augmentative communication systems have been excluded from educational and vocational training programs. In fact, Lindsay et al. (1986) reported the results of a demographic study indicating that only a very small proportion of the school programs developed for students using augmentative communication systems in the Province of Ontario were oriented towards "educational" goals of any sort. Rather these programs tended to emphasize behavioral management or therapeutic goals. Without access to education and vocational training, most augmentative communicators have faced considerable odds in maximizing their potential so that they can make a full contribution to society and enjoy a high quality of life.

To date, there have been no reports of research towards the needs of severely speech and/or writing impaired persons in prevocational or vocational settings (Beukelman, 1986). The requirements of the workplace impose unique communication demands for clients, including the need for telephone and computer access, demands for adequate rates of communication, etc. Research is required to assess the vocational and educational opportunities available to nonspeaking physically disabled individuals and to develop appropriate educational and vocational training programs to allow these individuals to meet the requirements of potential employment opportunities. Research is needed to consider strategies for keeping augmentative communicators within the educational curriculum as much as possible (personal communication, D. Beukelman, August, 1987) and to consider necessary curriculum adaptations to facilitate the learning process.

Recent technological advances have provided the potential for physically disabled individuals to achieve greater levels of independence, thereby increasing their educational and vocational opportunities. However, if these technologies are to be used effectively, research is required to identify the resources, training, and support required by educators and employers to successfully integrate the required systems into the school, home, and work place.

Perhaps one of the greatest barriers experienced by nonspeaking physically disabled individuals within educational and vocational settings is the limitation of their literacy skills (Kelford-Smith, Thurston, Light, Parnes & O'Keefe, 1987). Without functional literacy skills, it is difficult to achieve independence in daily living within today's print-based society. Research is required to identify the literacy requirements within educational, vocational, and community environments, to investigate the familial/ cultural and metalinguistic skills associated with the development of literacy, and to explore the most effective approaches to teaching nonspeaking physically disabled children to read and write.

Program evaluation

Too often the effectiveness of clinical, education, and vocational programs developed to meet the needs of nonspeaking physically disabled children and their facilitators is evaluated solely through the intuition and impressions of clinicians and educators. In order to ensure that these intervention programs are optimally effective, research is required to collect valid and reliable quantitative data from which to evaluate the outcome of intervention. The evaluation of assessment-intervention programs will allow clinicians and educators to determine if changes are occurring within client and/or facilitator skills and to evaluate if these changes are indeed the expected outcomes of intervention (Beukelman, 1986). There has been some initial research in the field to consider the outcome of intervention programs (e.g. Dowden, Beukelman, & Lossing, 1986; Glennen & Calculator, 1985; Reichle, Barrett, Tetlie, & McQuarter, 1987). Additional research is needed to determine successful interventions with clients and facilitators, to identify the need for modifications to intervention approaches so as to better meet the needs of various population groups, and to permit the refinement of specific intervention strategies. As Beukelman (1986) has suggested, "Communication effectiveness evaluation results will become the primary base of justifying and developing more appropriate interventions. In addition, the evaluations of effectiveness will play an increasingly important role in directing the modification and refinement of augmentative communication equipment" (p. 441).

Single case designs are particularly well suited to these research goals, since they allow flexibility of design, indepth analysis of variables, and identification of functional relationships between treatment and outcome variables. Furthermore, single case research designs accommodate the problems of a heterogeneous population and allow the evaluation of intrasubject and intersubject variability. In addition to short term research, there is also a need for longitudinal studies to explore intervention and the development of communicative competence over time. One of the impediments to evaluating intervention outcomes in the field of augmentative communication is the lack of criteria for evaluation and the dearth of psychometrically sound assessment tools. Research is required to determine appropriate criteria and tools for evaluation and to socially validate the focus, procedures, and outcome of intervention.

In order to ensure the accountability of service delivery programs, there is also a need for research at a broader macroanalytic level to evaluate the efficacy and cost-effectiveness of various models of service delivery and to identify the impact of these programs on a community, regional, provincial, national, and international basis. Most of the clinics authorized by the Assistive Devices Program of the Ontario Ministry of Health to prescribe augmentative communication systems for physically disabled children and young adults currently support extensive waiting lists. The delivery of clinical services for this population is impeded by the paucity of trained professionals with expertise in augmentative communication (Ontario Ministry of Health Assistive Devices Program, 1985) as well as by the lack of funding support for clinical service delivery. Impact studies will serve as a basis for assuring quality and efficiency of service delivery and for affecting public policy at the provincial and national levels of government and within the universities.

Summary of priorities for clinical research

The current base of empirically derived knowledge in the field of augmentative communication is scarce. As a result, clinical and educational programs are largely driven by the experience and intuition of the transdisciplinary team. Concerted research activity is required to develop a sound base of empirical knowledge to guide the decision making of clinicians and educators. Demographic research is required to identify the characteristics of the population of individuals who use augmentative communication systems, their communication needs, and the training and support required by their facilitators. This demographic research will serve to justify increased funding commitments to the field, to identify neglected segments of the population, to facilitate planning for effective service delivery to clients and facilitators, and to determine direction for future technical developments in the field. This research will serve to identify the training needs for professionals in a variety of disciplines and will provide a basis for planning preservice and inservice educational programs in the universities at the undergraduate, graduate, and post graduate levels.

At present, there is a critical need for research to consider issues related to the selection, customization, and integration of augmentative communication systems to meet the communication needs of nonspeaking physically disabled children. Specifically, this research should consider the user requirements imposed by various augmentative communication systems, both aided and unaided, light tech and high tech. It is only through a full understanding of the motoric, cognitive, linguistic, and sensory/perceptual skills required for effective and efficient system use, that clinicians will be able to optimize the match between user needs and skills, and available technologies. Assessment protocols and expert systems need to be developed to support clinicians in the complex process of system prescription and integration. Research is also required to consider the impact of augmentatitive communication systems on the user, on the partner, on the interaction process, and on social perceptions generally.

The mere provision of augmentative communication systems does not, in and of itself ensure effective use. It is therefore essential that future research is directed towards determining the knowledge, judgement, and skills required by individuals who use augmentative communication systems and by their partners to communicate effectively. This research will form the empirical base for developing more effective assessment- intervention programs for nonspeaking physically disabled children and their partners.

At present, many individuals who use augmentative communication systems have limited access to educational and vocational opportunities. Research is required to develop appropriate educational and vocational training programs to support these individuals in developing the skills to meet the requirements of potential employment opportunities. Special attention should be directed towards the development of literacy skills for without functional literacy skills it is difficult to achieve independence in daily living within today's information-based society.

As clinical, educational, and vocational programs are developed to meet the needs of nonspeaking physically disabled children and their partners, it is critical that on-going research be conducted to evaluate the efficacy and cost-effectiveness of these programs. This research will identify successful interventions for specific population groups and will indicate directions for program modification and refinement to ensure that clinical and educational interventions are truly contributing to the development of communicative competence by individuals who use augmentative communication systems.

Priorities for technical research and development

Recent technological advances have resulted in a proliferation of devices designed or adapted to meet the face to face and written communication needs of physically disabled individuals. There are, for example, close to 1,000 special adaptations, programs, and peripherals to allow disabled individuals to access computer technology (Brandenburg & Vanderheiden, 1986). Despite these technological advances, there are still many disabled individuals who are unable to access available technologies due to severe physical, cognitive, and/or sensory impairments. Furthermore, many of the individuals who are able to access available communication technologies have been frustrated by limitations such as the poor quality of synthetic speech and the reduced rates of communication compared to the rates of conversation and writing experienced by non-disabled individuals. As a result of these limitations, most nonspeaking physically disabled individuals have been unable to realize their full communication potential and to achieve a high quality of life. Clearly, further technical developments are required to better meet the needs of this population group.

To date, much of the technological development in the field has been led by what is technically possible and realizable, rather than by client needs and capabilities (Rothschild, Ryan, Parnes & Milner, 1987). It is essential to provide strong clinical leadership in the area of technical research and development by clearly articulating client needs and skills as the functional requirements for the development of augmentative communication systems. Consumer input to system development and evaluation is critical, for consumers themselves are the best judges of the strengths and constraints of various technologies.

Current priorities for technical research and development include the following:

1. research into the technical and functional integration of technology to meet the communication and daily living needs of disabled individuals

2. research to develop communication systems which provide greater flexibility in their design

3. research into innovative access methods to accommodate the needs and skills of users who are unable to achieve optimal access to current technologies

4. research and development to ensure that standard computer-based equipment is accessible to the population of disabled users, including research to overcome specific ergonomic barriers to full independence

5. research and development activities into acceleration techniques to permit disabled individuals to converse and write at rates which are comparable to those of non-disabled individuals

6. research into improved quality of speech output to provide greater intelligibility, to permit multi-lingual output, and to allow telephone access, and

7. research and development to allow access to telecommunications for nonspeaking physically disabled persons.

Each of these priorities is discussed in more detail in the following sections.

The technical and functional integration of technologies

Future research and development activities should consider the role of fundamental technologies (i.e. technologies which are not computer based) as well as advanced technologies (i.e. technologies which are computer based) in meeting the communication needs of nonspeaking physically disabled individuals. Too often it is erroneously assumed that advanced technologies are "better" than fundamental ones, such as communication displays, etc. Yet, as Vanderheiden (1987) has argued, "many of the problems faced by disabled persons are better solved through the use of fundamental technologies rather than through advanced aids" (p. 703). Future research needs to consider the interplay of these options and their respective strengths and constraints.

Most physically disabled individuals using augmentative communication systems use a variety of fundamental and advanced technologies to meet their communication, mobility, and daily living requirements. The challenge of providing such multi-component systems is one of successfully integrating the components for the client, both technically and functionally. Technical integration refers to the optimal mounting and positioning of the hardware in relation to the user, to other technologies, and to activities of daily living; functional integration refers to the client's appropriate and integrated use of various techniques within the natural environment. Tannock, Rothschild, and Beesley (1986) have documented the complexity of meeting the demands for technical and functional integration in a clinical case study of a physically disabled young adult. At present there are few guidelines for integrating multi-component technologies. Research is needed to explore the optimal approaches to technical and functional integration so as to maximize the client's independence and daily functioning.

Flexibility and simplicity in the design of communication devices

The early technical developments in the field of augmentative communication tended to be rigid in their design, offering fixed vocabulary options and organizations, and a limited array of input options and selection techniques. More recent developments have resulted in greater flexibility of design so that currently available communication devices allow for a variety of input devices (e.g. standard, expanded, or mini keyboards, single or multiple switch arrays, optical or lightbeam pointers), selection techniques (e.g. direct selection, scanning, and encoding techniques) and output modes (e.g. voice output, hard copy output, output on a screen or transient display), as well as the option of customizing the vocabulary set. While these devices offer more options than those previously available, they still fall short of the complete flexibility required to accommodate a full range of client needs and skills. Without full flexibility built into communication devices, the prescription of systems can not be completely customized on an individual basis to meet specific client requirements. Too often the user must accommodate to the system's requirements, rather than the system being customized to the user's needs and skills. (Coleman, Cook & Meyers, 1980). Research and development is required to ensure greater flexibility in system design to accommodate a full range of client needs and skills. As systems become available which allow such flexibility, research will also be necessary to develop assessment protocols and computer programs to support the transdisciplinary assessment team in determining user needs and requirements and fully customizing devices to optimize their potential

Too often greater flexibility of system design has resulted in greater complexity of design. Complex designs increase the "cost of learning" for clients, facilitators, and professionals in the field. Many technical developments in the field have served to by-pass physical limitations, but only at the expense of significantly increasing cognitive and sensory/perceptual requirements. For physically disabled individuals with concomitant cognitive or sensory/perceptual impairments, the increased demands of these technologies may exceed client capabilities. Future research and development should ensure greater simplicity in the design of the person-computer interface to reduce the cognitive and sensory/perceptual loads, to minimize the learning time required to achieve operational competence by the user, and to reduce the demands on facilitators and professionals to provide technological support to clients in the use of their communication systems.

Innovative access methods

As noted earlier, there are some nonspeaking physically disabled individuals who are unable to access currently available technologies due to severe physical limitations and/or concomitant cognitive, linguistic, and sensory/perceptual impairments. Research is required into innovative access methods to overcome the barriers experienced by these individuals. Current research and development is exploring the use of eye gaze systems and speech recognition systems as potentially viable channels of access to computer technology. While these developments seem promising, at present the technologies are very expensive and somewhat unreliable. Further research and development should be directed towards improving the available eye gaze and speech recognition technology and extending its application with the disabled population.

Gaddis (1982) has suggested that future research and development into computer access should also consider the potential of proportional controls and body-centered controls. Proportional controls can be used to generate a large number of selection positions with minimal movement, thus allowing greater sensitivity and accuracy of control for the physically disabled person. Body-centered controls are mounted directly on the user's body and may take maximum advantage of the client's reliable control acts. Body-centered controls "..bring the control to the person instead of making the person go to the control" (Gaddis, 1982; p.71). Research and development into innovative access methods will ensure that the population of disabled persons has efficient and effective access to augmentative communication technology as required.

Access to standard computer-based equipment

With rapid developments in the application of computer-based technology in today's society, the ability to operate computer technology is becoming a necessity if an individual is to participate fully in educational, vocational, community and home environments (Microcomputer Applications Programme, 1988). This technology may offer disabled individuals the opportunity to participate on an equal footing with non-disabled persons. However, as Vanderheiden (1985) has noted, the same computer-based technology has "the potential for becoming the greatest new handicap disabled persons will ever face" (p.1). Most standard computer equipment is being developed with little thought for the impact on the disabled population (Microcomputer Applications Programme, 1988). Although the rehabilitation field has tried to develop special interfaces and adaptive peripherals to allow disabled individuals to control personal computers, the developments in mainstream technology are so rapid that, as Vanderheiden (1983) has noted, the "retrofit" solutions to accessing problems developed in the rehabilitation field are obsolete within an exceedingly short time frame. In order to ensure that disabled individuals are not further disadvantaged by developments of mainstream high technology, future research and development should be directed towards developing computer standards for manufacturers to facilitate the accommodation of adapted peripherals and towards developing technical specifications to allow universal access for disabled and non-disabled persons. Initial work into computer standards is being conducted at the Trace Research and Development Centre at the University of Wisconsin-Madison, while initial research into the specifications for universal access is currently underway at the Microcomputer Applications Programme of The Hugh MacMillan Medical Centre in Toronto and IBM. Additional research is required to extend these initial efforts in the field. Specifically, research should address the ergonomic factors of computer access such as handling paper and disks and operating direct manipulation devices such as the mouse. At present, most disabled individuals are dependent on someone else to "hook up" their access system. Future research should consider the feasibility of remote connections, such as infrared connections, for input devices as well as further miniaturization so that devices are readily transportable (Shein, 1988).

Acceleration techniques to enhance the rate of communication

One of the most critical problems encountered by physically disabled individuals who use augmentative communication systems is the reduced rate of communication in daily interactions and in written communication (Foulds, 1987; Yoder & Kraat, 1983). The reduced rate of communication seriously affects the educational and vocational opportunities available and the amount and nature of interpersonal interactions possible. Recent technological advances have resulted in the development of various acceleration techniques which offer the theoretical potential to improve motor efficiency and to thus enhance the rate of communication. These acceleration techniques include linguistic prediction techniques and abbreviation expansion techniques. Linguistic prediction techniques include ones in which the computer system predicts the next letter or the entire word based on the letters already selected. Predictions may depend on the frequency of vocabulary used, the recency of use, and/or the grammatical construction of the client's message. Abbreviation expansion techniques can be used with words, phrases, sentences or longer text. These larger units are preprogrammed into the computer system and represented by a smaller set of keystrokes (i.e. numeric, alphabetic, or pictographic/iconic codes). To date, the clinical implications of acceleration techniques have not been fully explored. Light, Lindsay, Siegel and Parnes (1988) studied the information processing demands of various approaches to abbreviation expansion and concluded that the full potential of these techniques will only be realized if the cognitive demands to memorize codes are minimized so that using the codes becomes an automatic process. Further research is required to consider the human factors associated with acceleration techniques. As a result of this research, it will be possible to identify required modifications to existing techniques and to develop new techniques to maximize the efficiency of conversational and written communication by individuals using augmentative communication systems.

Improved speech output

The development of voice output communication aids has offered the potential for increased functioning for augmentative communicators by facilitating group interactions and interactions at a distance, by allowing greater independence in communication and greater conversational control, by providing a means to express emergency needs and wants, and by offering the potential to facilitate interactions between nonspeaking physically disabled children and their nonreading peers (Light, 1988b). Despite these potential benefits, currently available voice output devices suffer from serious limitations in their intelligibility. Results of research into the intelligibility of commercially available speech output systems suggests that, for many systems, intelligibility is by far inferior to that of natural speech (Mirenda & Beukelman, 1987). A new generation of speech output communication systems with improved intelligibility will soon be available to partially circumvent this problem. However, these systems are expensive and still lack access to the full array of paralinguistic and prosodic features which are so critical to conversational interaction. Research is required to further improve the quality of speech output and to ensure access to age and gender appropriate output with natural intonational features. Current limitations to the quality of speech output on communication devices have meant that most individuals using augmentative communication devices have limited access to telephone communication, yet telephone communication is critical in meeting vocational, personal, and daily living needs.

Although a few communication aids have been developed which provide output in other languages (Galyas, 1987), the majority of voice output communication aids have been limited to the English language. Given the cultural diversity within Canada, many nonspeaking physically disabled individuals require access to other languages to allow them to participate fully within educational, vocational, home, and community environments. Thus, research is required to develop multilingual communication aids which provide high quality speech output in various languages.

Access to telecommunications

Telecommunications are of critical importance to physically disabled individuals who may experience mobility restrictions within the community, school, and work place due to architectural barriers. Telecommunication networks provide a means to access friends, family, and the necessary services of daily living, (e.g. teleshopping, home banking). Computer-mediated telecommunications may also provide access to information services, user groups, databases, bulletin boards and library catalogues (Treviranus, 1988). For individuals who are housebound, telecommunications may provide the opportunity to perform computer-related work at home and to transmit the finished product to the employer through a modem (Treviranus, 1988).

Unfortunately, the area of telecommunications has largely been neglected in the field. Research is needed to develop techniques to allow full telecommunication access for physically disabled individuals who are unable to speak. This research should consider the special needs of persons who require telecommunication access, but are unable to read or write. At present, most communication systems provide output in alphanumerics only. Output in symbolic or pictographic systems is limited to specific applications. This limitation may be especially problematic for the user who is a nonreader and requires communication through a graphic system.

Summary of priorities for technical research and development

While the technical advances in the field of augmentative communication to date have been significant ones, there is a critical need for ongoing research and technical development to ensure that the next generation of communication aids meet the needs and skill requirements of nonspeaking physically disabled individuals more effectively and efficiently. This research should consider the role and interplay of fundamental and advanced technologies in meeting communication and daily living needs and should tackle the complex problem of integrating technologies both technically and functionally to maximize the independence and functioning of clients. Future research and development activities should provide greater flexibility of design so that communication systems can be fully customized to meet the individual client's needs and skills. This research and development should aim for greater simplicity in the design of the person-computer interface so that the cognitive and sensory/perceptual loads are reduced and the "cost of learning" for clients, facilitators, and professionals is minimized.

Since some individuals are unable to access the current generation of computer-based communication systems effectively and efficiently due to severe physical limitations and/or concomitant cognitive and sensory/perceptual impairments, research and development is required into innovative access methods such as eye gaze systems, speech recognition systems, proportional access techniques, and body-centered controls. Future research should also be directed towards developing computer standards and technical specifications for universal access to standard computer-based systems so that the mainstream technology in today's society (e.g. automated banking machines, office computer terminals) is fully accessible by the disabled population. Research should address the ergonomic barriers to computer accessibility including techniques to by-pass the handling of paper and disks, and the "hooking up" of adaptive peripherals.

In order to ensure that nonspeaking physically disabled individuals are able to converse and write at rates which are comparable to those of non-disabled persons, future research and development is necessary to explore more efficient acceleration techniques within computer-based communication devices. Research is also required to improve the quality of speech output in communication devices so that clients have access to highly intelligible, age and gender appropriate speech output which incorporates intonational and other paralinguistic features, which provides the option for multi-lingual output as required, and which supports easy telephone access. Research is also needed to develop techniques to allow full telecommunication access for physically disabled individuals who are unable to speak.

Within this paper, it has been possible to highlight but a few of the priorities for future technical research and development. As the field progresses and some of the current problems are solved, no doubt new priorities will emerge. As the future unfolds, it is essential to remember that technical developments should be driven by client needs and requirements. Consumer and clinician input to system development and evaluation is critical.

Conclusion

The field of augmentative communication has experienced tremendous growth in the past two decades offering the potential for increased independence and for improved access to communication for physically disabled individuals who are unable to speak and/or write. This potential will not be fully realized, however, without educational and clinical programs which are effective in providing the augmentative communication systems and skills training required by nonspeaking physically disabled individuals and their facilitators. In order to be optimally effective, clinicians and educators require access to sound, empirically based information to support their decision making roles in the assessment-intervention process. Thus, concerted clinical research is required to develop the knowledge base in the field. Technical research and development is required to forge a new generation of technologies to meet the communication needs of nonspeaking individuals more effectively.

The commitment, cooperation, and participation of a wide range of persons is required to make the necessary clinical and technical research a reality. Thus the challenges of the future are many:

- to consumers, families, clinicians, and educators to identify the issues of central concern and the problems to be addressed by researchers

- to researchers to develop appropriate methodologies to investigate these problems, to interpret the results, and to inform clinicians and educators of findings in order to improve the effectiveness of intervention programs

- to technical personnel to translate specifications of client needs and requirements into augmentative communication systems which are more effective and efficient;

- to professionals within other fields (e.g., psychologists, vision experts, social workers, etc.) to share their expertise and resources with professionals in the area of augmentative communication;

- to the universities to develop undergraduate, graduate and post-graduate level programs to train clinicians, educators, and researchers with expertise in augmentative communication;

- to clinical centres and educational programs to provide the administrative support for on-going research in the field; and

- to funding agencies to provide the financial support to allow clinical and technical research incentives in the area of augmentative communication to become a reality.

References

Aiello, S.C., (1980). Non-oral communication survey: A one-county assessment and demographic study. Unpublished manuscript, Plavan School, Fountain Valley, CA.

Beukelman, D., (1986). Evaluating the effectiveness of intervention programs. In S.W. Blackstone (Ed.). Augmentative communication: An introduction (pp.423-455). Rockville, MD: American Speech Language Hearing Association.

Beukelman, D., Yorkston, K. & Lossing, C. (1984). Functional communication assessment of adults with neurogenic disorders. In A. Halpern & M. Fuhrer (Eds.) Functional assessment in rehabilitation (pp. 101-115). Baltimore, MD: Paul H. Brookes Publishing Co.

Brandenburg,S. & Vanderheiden, G., (1986). Rehab/Education resources book series: Vol. 1. Communication aids; Vol.2. Switches and environmental controls: Vol.3. Hardware & Software, San Diego, CA: College Hill Press.

Buzolich, M.J., (1984). Interaction analyses of augmented and normal adult communicators. Unpublished doctoral dissertation. University of California, San Francisco,CA.

Buzolich, M.J., & Higginbotham, D.J., (1985). Analyzing and facilitating the communicative competence of augmentative system users. Short course presented at the Annual Convention of the American Speech Language Hearing Association, Washington, D.C.

Coleman, C.L., Cook, A.M., & Meyers, L.S., (1980). Assessing non-oral clients for assistive communication devices. Journal of Speech and Hearing Disorders, 45,515-526.

Coxson, L., & Mathy-Laikko, P., (1983). Listener reactions to three nonvocal communication outputs. In A.W. Kraat (1985). Communication interaction between aided and natural speakers: An IPCAS study report. Toronto: Canadian Rehabilitation Council for the Disabled.

Doherty, J., (1985). The effects of sign characteristics on sign and retention: An integrative review of the literature. Augmentative and Alternative Communication. 1, 108-121.

Dowden, P., Beukelman, D., & Lossing, C., (1986). Serving nonspeaking patients in acute care settings: Intervention outcomes. Augmentative and Alternative Communication. 2,38-44.

Dowden, P., Honsinger, M., & Beukelman. D., (1986). Serving nonspeaking patients in acute care settings: An intervention approach. Augmentative and Alternative Communication 2, 25-32.

Foulds, R., (1987). Guest editorial. Augmentative and Alternative Communication, 3,169.

Fristoe, M., & Lloyd, L., (1978). A survey of the use of non-speech systems with the severely communication impaired. Mental Retardation 16,99-103.

Gaddis, E.L., (1982). Proportional computer interface increases speed of non-vocal communication. In Proceedings of the IEEE Computer Society Workshop on Computing to Aid the Handicapped, (pp. 69-75). University of Virginia, Charlottesville, VA.

Galyas, K., (1987). Voice output communication aids: A survey and an evaluation. In Proceedings of the RESNA 10th Annual Conference (pp. 97-99) San Jose, CA. Glennen, S. & Calculator, S. (1985). Training functional communication board use: A pragmatic approach. Augmentative and Alternative Communication 1, 134-142.

Kelford-Smith, A., Thurston, S., Light, J., Parnes, P., & O'Keefe, (1987). The Form and use of written communication produced by physically disabled individuals using microcomputers. Manuscript submitted for publication.

Kraat, A.W., (1984). Communication interaction between aided users and natural speakers - An international perspective. Proceedings of the Second International Conference on Rehabilitation Engineering (pp. 43-46). Ottawa, Canada.

Kraat, A.W., (1985). Communication interaction between aided and natural speakers: An IPCAS study report. Toronto: Canadian Rehabilitation Council for the Disabled.

Light, J., (1988a) Interaction involving individuals using augmentative and alternative communication systems: State of the art and future directions for research.

Light, J.,(1988b) Augmentative communication: State of the art in North America. Paper submitted to the International Conference of the Association for the Advancement of Rehabilitation Technology, Montreal, Canada.

Light, J., Collier, B., & Parnes, P., (1985). Communicative interaction between young nonspeaking physically disabled children and their primary caregivers: Part III - Modes of communication. Augmentative and Alternative Communication, 1,125-133.

Light, J., Lindsay, P., Siegel. L., & Parnes, P. (1988).Information Processing demands of message encoding techniques. Manuscript submitted for publication.

Lindsay, P., Cambria, B., & McMaughton. S., (1986). The educational needs of nonspeaking students and their teachers or primary caregivers. Paper presented at the Fourth Biennial International Conference on Augmentative and Alternative Communication. Cardiff, Wales.

Matas, J., Mathy-Laikko, P., Beukelman, D., & Legresley, K., (1985). Identifying the nonspeaking population: A demographic study. Augmentative and Alternative Communication 1,17-31.

Mathy-Laikko, P., & Yoder, D., (1986). Future needs and directions in augmentative communications. In S.W. Blackstone (Ed.) Augmentative Communication: An introduction (pp. 471-494). Rockville, MD: American Speech Language Hearing Association.

Microcomputer Applications Programme, The Hugh MacMillan Medical Centre (1988). Selecting access systems for persons with physical disabilities. Resource book (under development)

Milner, M., & Parnes, P., (1984). Development and evaluation of a multi-faceted communication tray for vocabulary expansion. Unpublished manuscript, The Hugh MacMillan Medical Centre, Toronto, Canada.

Mirenda, P., & Beukelman, D., (1986). A comparison of speech synthesis intelligibility with listeners from three age groups. Augmentative and Alternative Communication, 3,120-128.

Ontario Ministry of Health Assistive Devices Program - Communication Aids Subcommittee (1985). Paucity of appropriately trained professionals with relevant skills in disciplines relating to the provision of communication aids via the Assistive Devices Program.

Prutting, C., & Kirchner, D., (1987). A clinical appraisal of the pragmatic aspects of language. Journal of Speech and Hearing Disorders, 52,105-119.

Reichle, J., Barrett, C., Tetlie, R., & McQuarter, R., (1987). The effect of prior intervention to establish a generalized requesting on the acquisition of object signs. Augmentative and Alternative Communication, 3,3-11.

Rothschild, N., Ryan, S., Parnes, P., & Milner, M., (1987). Collaboration - An essential issue in the development of Augmentative communication devices. In the Proceedings of the RESNA 10th Annual Conference (pp.91-93). San Jose,CA.

Shein, F., (1988). Future directions of computer access technology for physically disabled persons. Unpublished manuscript, The Hugh MacMillan Medical Centre, Toronto, Canada

Tannock, R., Rothschild, N., & Beesley, M. (1986). Integration of multiple communication devices: A case study and inception of a model. In Proceedings of RESNA 9th Annual Conference (pp. 307-309). Minneapolis, MN.

Treviranus, J., (1988) Telecommunications and the augmentative communication user. Paper submitted to the International Conference of the Association for the Advancement of Rehabilitation Technology, Montreal, Canada.

Vanderheiden, G., (1983). The practical use of microcomputers in rehabilitation. Rehabilitation Literature 44,66-70.

Vanderheiden, G., (1987). Service delivery mechanisms in rehabilitation technology. The American Journal of Occupational Therapy, 41, 703-710

Vanderheiden. G., (1985). Access to standard computers, software, and information systems by persons with disabilities. Report on the Proceedings of the Meeting of the Government Industry Initiative on Computer Accessibility by Individuals with Disabilities.

Vanderheiden, G., & Lloyd, L.,(1986). Communication systems and their components. In S.W. Blackstone (Ed.). Augmentative Communication: An introduction (pp.49-161). Rockville M.D.: American Speech Language Hearing Association.

Warrick, A., (1988) Sociocommunicative considerations within augmentative and alternative communication. Augmentative and Alternative Communication, 4,45-51.

Yoder, D., & Kraat, A., (1983). Intervention issues in nonspeech communication. In J. Miller, D. Yoder & R. Schiefelhusch (Eds.). Contemporary issues in language intervention. ASHA Reports 12 (pp. 27-51). American Speech Language Hearing Association, Rockville, MD.

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Priorities for research in the field of augmentative communication

Summary

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The development of fundamental and advanced technologies in augmentative communication paired with developments of other assistive devices has provided the potential to significantly reduce the disability and concomitant handicaps experienced by many physically disabled individuals. To date, there has been only minimal research into the myriad issues involved in the application of these technologies. There is a critical need for concerted research to develop a sound empirical and theoretical base of knowledge to guide clinical practice and to specify the requirements for technical research and development into new generations of communication systems and other supporting technologies to better meet the needs of disabled individuals.

Within the province of Ontario, we are fortunate to have the support of the Assistive Device Program of the Ontario Ministry of Health. Through the authorized clinics within this program, physically disabled children and young adults are provided with access to the augmentative communication systems and other assistive devices (e.g., mobility aids, sensory aids, etc.) which they require to achieve their full potential. In setting priorities for research in childhood physical disabilities within the province of Ontario, it is important to consider not only the questions which appear to be of primary interest, but also the strengths and resources available within the Province. In other words, it is important to consider what it is that researchers in the province can do to make the most meaningful contribution within the field at provincial, national and international levels. The Assistive Devices Program and the clinical network in the province provide an ideal support system to allow us to take a leadership role in research in the application of augmentative communication systems and other assistive devices.

In order to assume such a leadership role, however, we require support in the following areas:

1. Program funding to allow for the development of centres of excellence having comprehensive and committed research teams with links to clinical/educational settings to explore the complex research issues which require long term investigations (5-10 years). These centres of excellence would provide leadership in the field, would have the flexibility to generate innovative ideas for the future, and would offer the required expertise to foster young researchers in the field of (re)habilitation.

2. On-going project funding to allow researchers to address the more specific questions which require short term funding (2-3 years). Project funding will provide solutions to today's problems and will contribute knowledge to the field as a foundation to future developments.

3. Support for new researchers to develop expertise within the field. There is currently a paucity of trained professionals in the field of augmentative communication and assistive devices, especially research personnel. The research problems confronted in (re)habilitation are complex ones. Researchers are required who have the necessary expertise, enthusiasm, and creativity to meet the challenges posed. Stronger links need to be forged between the clinical/educational centres and the institutes of higher education in the province so that students at the undergraduate and graduate levels develop greater knowledge and interest in childhood physical disabilities.

4. Support for networking and collaboration amongst research teams. As Alan Newell noted in his paper, links should be promoted amongst groups to encourage a rich exchange of ideas and directions. These links might be promoted through visiting scholar programs, clinical and research sabbaticals, and international "think tanks" and symposiums.

With the support outlined, we will be able to assume a strong leadership role in the area of augmentative communication and other (re)habilitation technologies. Specific research priorities within the field of augmentative communication have already been articulated with the papers presented by Alan Newell and myself at the conference. To summarize briefly, priorities in clinical research include the following:

1. Demographic research to identify the needs and characteristics of individuals requiring augmentative communication systems, as well as the needs and resource requirements of their facilitators

2. Research to guide in the selection, customization and integration of augmentative communication systems

3. Research to identify factors which contribute to the development of communicative competence

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4. Research to delineate the requirements for educational curicula and to determine vocational opportunities; and

5. Research to evaluate the efficacy and cost effectiveness of intervention programs.

Priorities for technical research and development include the following:

1. Research into the technical and functional integration of technology

2. Research to develop communication systems with greater flexibility and simplicity in their design

3. Research into innovative access methods

4. Research and development to ensure that standard computer-based equipment is accessible

5. Research into acceleration techniques

6. Research into improved speech output; and

7. Research and development to allow access to telecommunications.

Within all of these research efforts, as Alan Newell suggested, we must encourage a thorough knowledge of current practices, but we must also promote creative and innovative approaches and the generation of "novel" ideas.

Reducing the disability - communication technology

Discussion

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I am pleased to be a participant in this International Conference on Priorities in Childhood Physical Disability and I am especially pleased to have the opportunity to discuss the two papers that you just heard regarding research priorities in this area. These two authors, Light and Newell, have presented a lot of material, with each author having selected a different approach to the subject, Newell's paper addressing priorities at a more global level and Light's paper at a more specific level. Consequently, you have had painted for you both the large research picture frame and also the more detailed subject matter within it. To illustrate the picture, how can technology for the disabled person be improved as a result of fostering research in general and further, what specific programs are needed for our researchers, scientists, and professionals to carry out to reduce the disability of physically disabled children? Whether these different approaches emerged by design or serendipitously, they have the marvellous result of providing a very broad scope of material to digest. As a discussant, however, covering all the issues raised and addressing each at both a general and specific level is not possible in the time available. Therefore, my discussion will be selective and highlight only some issues.

First, I will discuss some global issues. Newell mentioned the need to create a research infrastructure and atmosphere that would encourage new ideas, which, if created, would require a long-term time frame, such as five to ten years. Productive research to solve complex problems indeed requires long-term goals. In technology, this is equally true. For example, a recently initiated research project at the Kluge Children's Rehabilitation and Research Institute in Charlottesville, Virginia has adopted the goal of developing a device that will produce speech in real time. The time frame to achieve this goal is ten years. Newell also commented that a long-term goal is one of radical change rather than one of adopting a step-by-step pathway that ends at the achievement of a goal. What he is suggesting seems consistent with how science and research evolve. Research is not analogous to running a straight alley maze with a single path, which starts at A and ends at B. Rather research is more like solving a maze puzzle, with many blind alleys and circuitous paths en route to determining the shortest path to the goal? Therefore, we need to allow the time and support for researchers to pursue long-term goals. What does this mean pragmatically? I think that it means longer grant funding periods, increasing grants from one and two years to at least three to five years and preferably longer, where warranted. A positive step in this direction has been the initiation of the Javits Neuroscientist Investigatorships within the National Institute of Neurological and Communicative Disorders and Stroke (NINCDS) of the National Institutes of Health (NIH). These awards have been strongly endorsed among scientists since they provide funding for a seven year period. To reduce the application-progress report-reapplication paper cycle associated with continuity of funding, there is also a move to NINCDS to provide individual investigator research grant awards for three to five year periods. To achieve long-term goals funding must support both the up-and-coming researchers, those with great potential, as well as the successful scientists with already established careers. An evaluation of NIH's Research Career Development Award (RCDA) program by the Rand Corporation revealed that the awardees were successful scientists with very productive research careers. However, the awards tended to be made to the already successful rather than to the target group the prospectively successful (Carter, Winkler, & Biddle, 1987).

If radical change is necessary, what will bring it about? In addition to the time frame mentioned above, Harvey Long (1988) from IBM addressed this issue in a recent panel discussion at a conference entitled "Technology; Advancing Your Profession." Long indicated that in times of rapid change your worst enemy is experience. You need to get around complacency and job security. As Long put it: "Evolve or dissolve."

I believe that all of us would agree with Newell's recommendation of the need for a multidisciplinary research team. He also mentions the need for a more active role by speech-language pathologists. Communication is our area of expertise - we must play a major role if we are to assume our full responsibilities as professionals. Our role should encompass not only clinical service provision to alternative and augmentativecommunication device users but also utilization of our knowledge of speech and language science in the design of input and output components of these systems as well as their language content.

Developing partnerships is a need in augmentative communication that I believe can help to achieve our set goals (Shewan, 1988). That means linking researchers with one another using mechanisms, such as telecommunications (SpecialNet, CONFER, etc. and other bulletin board, electronic mail, and conferencing systems) and face-to-face conferences which promote exchange and cross-fertilization of ideas. Among the foundations that sponsor and support such activities are the National Science Foundation, the Rockefeller Foundation, and the American Speech-Language-Hearing Foundation. Annual meetings of organizations such as ISAAC also help to meet this need. Partnerships are also mutually beneficial among industry, professionals, and universities.

Light's paper provided an excellent coverage of the types of research needed clinically and technologically, some of which I will highlight. In addition to these two kinds of research, however, a third kind of research has the potential for contributing to the area of alternative and augmentative communication. I am referring to basic research, that is, research not targeted to a particular end product, such as an augmentative communication device, but rather to research to advance our knowledge bases of speech and language processing, cognitive processing, social interaction, etc., in general. From this evolving new knowledge may emerge applications to augmentative and alternative communication. For example, NASA did not set out to design many products that we take advantage of and use daily. However, the goal of putting a person on the moon had the ultimate effect of creating many by-products that we now enjoy. So I would like to add to the research priorities the support of basic research as a necessary undergirding for knowledge advancement. Of course, specific research programs designed to enhance augmentative and alternative communication are equally important. The need for clinical or applied research and for better quality research has been the subject of many papers, Light's included. As she pointed out, research is needed to guide the decision making of clinicians and educators, to examine the efficacy of our service delivery programs, to delineate requirements for educational criteria, and to guide the selection of augmentative communication systems. How do we get more research and better research? The path seems to lead back to fostering research in our educational preparation programs to prepare our students to carry out research as an integral part of their professional lives. For this to occur in augmentative communication, university training programs need to teach students about the area. As late as November 1984, only 46% of speech-language-hearing graduate curicula provided a separate course in augmentative communication and an additional 14% provided augmentative communication as part of another course. That more experienced professionals were needed to provide the teaching in this young field led to the conduct of a "Leadership training in Augmentative Communication" grant funded by the U.S. Department of Education and carried out by the American Speech-Language-Hearing Association (ASHA, 1986). Of additional encouragement is the recent development of three doctoral programs with specialties in technology/communication disorders (John Hopkins University, Purdue University and the University of Nebraska). Professors who are doing the teaching in augmentative communication also need to be actively engaged in research in this area to provide research role models, practical experience, and to foster critical thinking skills necessary for good research. More programs like the National Science Foundation's Research for Experiences for Undergraduates (1987) should be initiated. For those already practicing in the field, there is a need to acquire research skills through continuing education programs assisting in research project, etc. In these ways, current and future professionals will have the role example that research can be and is a part of being a professional.

Important to emphasize are the needs for prevention, early identification, and treatment that is efficacious, effective, and efficient. It has been well documented in the literature on hearing impairment and special education that early identification is important to reduce the degree of disability (Asha, 1988; California State Department of Education, 1987). In addition, early special education intervention has been demonstrated to be cost effective and efficacious (California State Department of Education, 1987, Garland, Stone, Swanson, and Woodruff, 1981). We share these needs with other disciplines, such as learning disabilities, and they are among the major foci of my profession, the speech-language-hearing profession. Progress is being made, although we have a long way to go. Assessing the effectiveness of clinical procedures appears as a highest priority objective in ASHA's 1988-1990 long range plan and research on the detection, prediction, and prevention of speech, language, and hearing disorders and research of the efficacy of intervention are two of the three research priorities adopted by ASHA's Committee of Scientific Affairs. Legislatively, Public Law 94-142 has mandated special education services for infants and toddlers (0-2 yrs.) and has expanded special education services for 3-5 year olds in the schools. As consequence, the Department of Education, through the Office of special Education and Rehabilitation Services (OSERS) has included in its 1988 priorities programs dealing with, (l) developing new and improved procedures in serving infants and toddlers with specific disability conditions and (2) research projects that compare compensatory strategies that result in functional skills, such as the use of augmentative communication systems, or other types of adaptation, or technological applications that enable functional responding. While initiatives are being put forward, great effort is needed if we are to achieve the goal of research-based clinical decisions.

Another research area necessary if we are to provide services effectively is that of how augmentative communication affects the development of the child. Of special concern is how an augmentative communication system affects the development of literacy skills - reading, writing, and spelling. Do the devices, such as MINSPEAK have a positive, negative, or neutral influence? What is the effect of programs like SPELLWRITE by Beukelman on the spelling skills of augmentative communication system users? Dede (1988), citing the activity of composing a sentence, has noted that different skills are required and that the ways we accomplish tasks are different when using a computer versus pencil and paper. We can extend this concept to augmentative communication. As Vanderheiden and Yoder (1986) and Light have noted, facilitators need to know what skills and processes are involved in operating an augmentative communication device, such as a computer, and here we can not use the normal acquisition model. We have to specify these skills from research with augmentative communication system users, so that we can apply the knowledge to new users. If you want to know how aphasic patients perform, it is best to observe aphasic patients and not normal language users. With the advent of expert systems and computers that can mimic the brain activity patterns of primates engaged in cognitive tasks (The Chronicle of Higher Education, March 2, 1988) perhaps our future research can show us how to adapt augmentative communicative systems to transform language processing tasks in ways so that our augmentative communication system users can accomplish them.

To be maximally useful, augmentative communication systems can not be static systems. They must keep pace as a client changes in his/her life path; with children it is hoped that these changes will be in a developmental direction (ASHA, 1987). We must develop multi-component systems that address the many functions in communication, both now and in the future (Vanderheiden and Lloyd, 1986; Yorkston and Karlan, 1986). We need research that tells us how to meet consumer needs, to use Newell's expression. To do that we must address the augmentative communication system user, the device used, the communication partners, and communication environments, as Light has mentioned. Blackstone (1987), in her final grant report entitled "Implementing Strategies for Improving the Use of Communication Aids in Schools Serving Handicapped Children," grouped such needs into four broad areas; professional competencies, service delivery, quality assurance, and client intervention. Because augmentative communication use requires different skills from regular oral and written communication, we need to develop both assessment tools that measure the skills required and a different frame of reference. In order to lead lives that are fully integrated in the community, the augmentative communication system must be functional in many environments - home, work, recreational, and social settings. Research is needed to determine what skills will be required by the augmentative communication system user and how to teach them.

Among the technological priorities mentioned by Light were research to develop better speech recognition and better voice output and augmentative communication devices that were more communication competence oriented. Recent developments have brought us to the era of conversational computers. Poock (1988) demonstrated conversational speech recognition computers that, although requiring good training, are very accurate in transcoding from speech to text. They can decode series of numbers or sentences and can account for changes in speech due to emotional, mental, or physical stress. Tangora is an IBM system that has developed an experimental, large vocabulary, isolated word speech recognition system whose technology has applicability to people with different types of handicaps.

Speech production has also dramatically improved in 1988. DEC Talk is now available in a portable form. we are turning the corner with synthesized speech and developing devices that can achieve the minimum 81% intelligibility level. The diphone method, which utilizes the transitions between phonemes in its algorithms, represents another improvement in intelligibility of synthesized speech. One product using this method is REALVOICE (Blackstone, 1988). Schmandt (1988) computer system that gives directions to a driver. It uses knowledge of rules of discourse (acknowledgement, pause, repeat, etc., to manage the discourse flow control while presenting driving instructions with synthesized speech. With more research, these systems will potentially be able to operate in more varied environments.

The output of augmentative communication devices should not be limited to speech alone. Recently, Romich (1988) demonstrated singing on a computer. This development arose from a need expressed by augmentative communication system user children attending a camp. This is a good example of augmentative communication driving technology and not the other way around. According to Romich, technology is not driving the augmentative communication applications, rather augmentative communication needs are requiring the development of technological changes. Hopefully, this work will also extend to meet the need addressed by Light for multilingual systems.

The key to being up-to-date in the future will hinge on knowing how to get information. With computers and augmentative communication systems this means keyboarding skills and access to data bases. Therefore, ways to access information are crucial to augmentative communication users. Research is needed, with one of the exciting places undertaking it being the Trace Research and Development Centre at the University of Wisconsin-Madison. This facility was recently awarded the Rehabilitation Engineering Centre for Access to Computers and Electronic Equipment by the National Institute on Disability and Rehabilitation Research. (NIDRR). As many of you know, Greg Vanderheiden is director of the Trace Centre. Another Rehabilitation Engineering Centre for Augmentative Communication was awarded to the University of Delaware, with Rick Foulds as director.

1988 finds us having made great strides in augmentative communication. However, the research base is small and not of optimum quality. There is a need both more and better research as Newell and Light's papers have outlined in more detail. we need to gather data to make reasoned decisions about augmentative communication systems, users, and their interface as well as to explore the future horizons of technology for its benefits to augmentative communication users. The road of influence between augmentative communication and technology must run in both directions, to the mutual benefit of both areas.

References

American Speech-Language-Hearing Association. (1986). Leadership training in augmentative communication. Final Report. Department of Education, Special Education Programs, Personnel Preparation Award Number 84 029.

American Speech-Language-Hearing Association. (1988). Guidelines for the identification of hearing impairment in at risk infants age birth to 6 months. Asha, (4), 61-64.

American Speech-Language-Hearing Association. (1987). Implementation Strategies for Improving the Use of Communication Aids in Schools Serving Handicapped Children. Final Report. Department of Education Special Education Programs, Contract No. 300-885-0139.

Blackstone, S.W., (Ed.) (1986). Augmentative communication; An introduction. Rockville, M.D., American Speech-Language-Hearing Association.

Blackstone, S.W., (Ed.) (1988). Augmentative communication news. 1 (1) , 1-8.

California State Department of Education. (1987). The efficacy and cost effectivenes, of early education for handicapped infants and preschool children. Sacramento, CA.

Carter, G.M., Winkler, J.D., & Biddle, A.K., (1987). An evaluation of the NIH research career development award. Santa Monica, CA, The Rand Corporation.

Computer Model of Brain's Activity. (1988). The Chronicle of March 2.

Dede, C.J., (1987). Empowering environments, hypermedia and microworlds. The Computing teacher, November, 20-24.

Garland, C., Stone, N.W., Swanson, J., & Woodruff, G., (Eds) (1981). Early intervention for children with special needs and their families. Westar Series Paper #11.

Long, H., (1988). Expert Panel Presentation at the Technology, Advancing Your Profession Conference, Mesa, Arizona.

Poock, G., (1988). Conversational computer systems. Presentation at the Technology, Advancing Your Profession, Conference, Mesa, Arizona

Romich, B., (1988). Augmentative communication synthetic and digital speech, In perspective, in use and in the future. Presentation at the Technology, Advancing Your Profession Conference, Mesa, Arizona.

Schmandt, C., (1988), Conversational computer systems. Presentation at the Technology, Advancing Your Profession Conference, Mesa, Arizona.

Shewan, C.M., (1988), Computer manufacturers and professionals; Partners in research. Research Bulletin, April, 21-22.

Vanderheiden, G.C., & Lloyd, L.L., (1986). Communication systems and their components In

Sarah W. Blackstone (Ed.) Augmentative communication; An introduction. Rockville, M.D.; American Speech-Language-Hearing Association.

Vanderheiden, G.C.,& Yoder, D.E., (1986). Overview In Sarah Blackstone (Ed.) Augmentative communication: An introduction. Rockville, M.D., American Speech-Language-Hearing Association.

Yorkston, K.M., & Karlan, G., (1986). Assessment procedures. In Sarah W. Blackstone (Ed.) Augmentative communication: An introduction. Rockville, M.D., American Speech-Language-Hearing Association.

10. Functional improvement - environmental control and mobility

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Parenting and schooling are meant to facilitate the acquisition of knowledge, experience and skills by children that will lead to independence. Learning occurs by way of example, play, exploration and training. Young children learn that the majority of events happen for a reason and that they can control events in their environment. Children with physical disabilities often have limited opportunities to explore and learn from their surroundings. Control of one's environment can be achieved in one of two ways. Either a person can develop the skills needed to manipulate the conditions or elements which make up the surroundings, or the conditions or elements can be modified to match the person's abilities to direct them. Both of these approaches to environmental control for children with physical disabilities need to be explored. Consideration must be given to the home, school and community environments and to the need to be able to move with freedom within and between these environments. This paper primarily considers the role technology can play in providing access to the environment and draws to a large extent on projects that have or are in the process of evolving from the Hugh MacMillan Rehabilitation Centre.

The home environment

Environmental control systems within the home setting are provided for the operation of appliances and/or the telephone. Access to the systems may be via a variety of input methods such as a "sip and puff" interface that may be encoded to operate a number of different functions, capacitive or other types of contact switches (Symington et al, 1980) and voice recognition (Snell et al, 1987). Operations requiring activation of a switch such as turning on and off a radio, light, fan and stereo, dialing a number or answering the telephone, opening doors, and changing channels on a TV or adjusting the volume, can be controlled through the environmental systems. Most such systems are used by adults who have a high level spinal cord injury.

Integrated Systems

The advent of affordable personal computers has led to the development of integrated systems that can also be used to run commercially available software on the computer. For example, a person can have access to word processing or spread-sheet programs either for correspondence and to manage personal finances, or to gain employment. An important attribute of such systems is that the majority of parts are "off-the-shelf" so that they are more affordable than equivalent devices developed specifically for the disabled population. Furthermore, system components can he serviced through commercial houses which are more widely available than specialised rehabilitation centres. A disadvantage of present systems is that the user must be in close proximity to operate them. The systems need to be able to operate remotely in closed loop fashion i.e. the systems should be able to acknowledge what instructions they have received for execution.

A similar approach has been taken in providing non-speaking children with communication systems. If a child has a physical disability that limits control to activating one or two switches, the action of the switches can be used to highlight words arranged in a matrix on a communication board. The action of the switches can be combined with a scanning strategy that highlights elements, rows or columns of the matrix in turn. A switch can be activated to initiate the scan or the system can scan automatically. Another switch would be used to change from column to row scanning, say, by selecting a particular column. Lastly, the desired element within the column would then be selected. The item selected could be a symbol or word to be communicated or it could be an instruction to an environmental control system or educational computer program. The means of access would be the same in all cases. Only the application would differ. However, the power of evolving computer systems, some of which are portable, should be used to accommodate all three applications. Implementation of such a system is technically feasible. The challenge facing rehabilitation professionals is ensuring the wise prescription of such a system. Multidisciplinary teams first need to assess the abilities and disabilities of the individual client and identify his or her needs. A program of achievable goals should be set out for the client that accounts for any accompanying sensory, cognitive or perceptual deficits. Reassessments should occur regularly so that timely changes to the program can be made. Training, because of its labour intensive nature and long duration, should be undertaken primarily in the home or residence environment.

Training

One approach could be to create a computer-based training and assessment package that will place minimal requirements on the parent or caregiver. For example, the training system could be used to provide the client with an understanding of the cause-effect relationship that exists between the activation of a switch and its consequence, the operation of a toy. Information regarding numbers and timings of switch activations could be downloaded to the rehabilitation centre via modem. The information could then be used to decide on whether to modify or maintain the current training regimen. Changes in regimen could be conveyed, again via the telephone lines, from the rehabilitation centre to the computer to modify the training regimen. The current move towards deinstitutionalization of persons with disabilities means that family members and local community services will be required to provide increased care. Automated remote training and assessment along the lines described should considerably facilitate care-giving by eliminating the need for frequent travel to the rehabilitation centre.

Adaptive Filtering

If a child is able to grasp a controller such as a joystick and is able to understand how it can be used to control devices, then technology can be used to compensate for any unwanted motions imposed upon purposeful input. Traditional engineering approaches such as the use of self-adaptive proportional-integral- differential filters have been used in the past to adjust wheelchair performance to variations in load. However, parameter variations resulting from fatigue or varying degrees of athetotic movements can alter the system model characteristics from moment to moment. The potential exists to create rule-based control systems that can develop a qualitative cause-effect model of the person who is using the device, and learn rules on-line as time progresses. Such systems, if implemented, should be able to compensate for changing physical abilities in persons with progressive disease such as muscular dystrophy or amyotrophic lateral sclerosis without requiring modification. For example, as a person's ability to produce force on a proportional joystick decreased over time, the system would automatically monitor this and adjust for it. Furthermore, such intelligent systems could be used to record a client's changing physical ability over time and make such information available to the prescribing rehabilitation team.

Sound Recognition

Voice recognition as an input modality to a computer-based environmental control system has been implemented and successfully used. Most voice recognition systems are, in effect, sound recognition systems, that is, they can be trained to recognise a series of distinct sounds. The use of such systems with children with cerebral palsy who are only able to utter sounds needs to be explored. It may be possible to recognise a number of distinct utterances for an individual that can be repeated with sufficient reliability so that sound could be used to simulate a single or multiple switch input to a control system.

Metabolic Cost

Often children with physical disabilities can directly manipulate objects that have been modified to facilitate handling. An argument can be made that low technology devices are economical, reliable, and bring the child in contact with the "real" world. However, at times the effort needed to manipulate devices is sufficiently demanding that the child becomes unable to maintain an activity. The area of metabolic energy expenditure is one that requires further investigation so that guide-lines can be established when, for example, to provide a child with a powered wheelchair because if a manual wheelchair is used, the child, having arrived at his or her destination, is too tired to do anything.

Robotics

Robots have been investigated as a means whereby disabled people can manipulate objects through remote control. A front-end voice recognition system or joystick are usually employed by the operator to convey commands to the robot.

Research is presently underway in a number of centres to develop control strategies that incorporate preprogrammed tasks that will allow the robot to approach the vicinity of the target with a minimum number of commands. Feedback through pressure switches and sight systems is being explored as ways of improving robot performance.

However, the skills needed to operate such systems will preclude many children from eventually being able to use them. One approach presently under consideration (Halpern, 1988) to overcome this problem uses a computer to simulate the robot's environment. The user, who may be remote from the mobile robot, carries a graphical representation of what is before the robot on a video screen. By manipulating a cursor on the screen and activating an action switch, the user can direct the robot to the fridge, say, extract a plate and place it in a microwave oven, set the keypad of the microwave oven, and then have the heated plate carried to where the person is. The view presented to the person will always graphically depict what the robot is "seeing" so that the person will at all times feel that he or she is in control.

The school environment

In the previous section, mention was made of how computers can be accessed through as little as a single switch. In fact, a variety of adaptations exist that can be used to facilitate computer access. These include head sticks, keyboards of various dimensions, keyguards, visual keyboards accessed by combinations of switches and scanning strategies, optical or ultrasonic headpointers, joy sticks and trackballs. In the past, dual computer systems have been developed. The "front-end" computer is used as the person's access method to the "back-end" computer. The latter receives commands and interprets them as though these are entered directly through the keyboard to run unmodified, commercially available software. The visual keyboard can consist of traditional orthographic symbols or some other representation such as Blissymbols. Another approach has been the use of a transparent keyboard in the form of a pop-up window along the bottom of the screen. Selections are sent directly to the keyboard buffer of the computer. With the advent of multitasking, it will now be possible to create and rapidly display a general selection set (visual keyboard) or selection sets that are specific to an application program. The effect on the user of positioning of the selection set window with respect to the application program window and the size of its elements are yet to be assessed. Nevertheless, the flexibility of computers provides means whereby access to educational software can be tailored to the individual once guide-lines for interfacing have been established.

Educational software should be used to create an environment that will allow students to explore and develop an understanding of the subject matter, progressing from simple to complex facts, relationships and skills. For example, in teaching physics, the computer can be used to simulate models of rules and relations that govern the movement of objects, work done, energy and the effect of friction or mass. A kit that could accompany the simulation program might consist of radio controlled battery powered cars of different weights and performance characteristics, two road surfaces with different friction coefficients, and risers, starting gates and a mechanism for measuring distance. The complete set up should be operable through the computer and accompanied by appropriate support text for the teacher and pupil. Systems such as the one described already exist and allow both physically disabled and able-bodied students to manipulate objects both in a concrete and simulated form, albeit in a remote manner. This approach to teaching will allow each child to proceed at his or her individual rate of learning while providing for a range of learning outcomes.

The more opportunity children have to experience, explore and affect their environment, the more chance they have of integrating concepts, compensating for disabilities, and choosing alternative means of functioning in their environment. The hypothesis is that an environment that facilitates, makes accessible and simulates the ordinary experience of milestones for children who are unable to otherwise experience them, will enhance, reinforce and improve their cognitive abilities. For the young child classrooms should be designed to maximize the accessibility of play areas. If the child is unable to move through, in and around the spaces, the child could use remotely controlled robots to explore the environment and gain experience with directionality and spacial relationships while promoting spontaneous interaction with other children. Methods are needed to assess the effect of such environmental changes on the child's development and refine the design of such play areas. The need for such adaptations and their evaluation will become more urgent as mainstreaming is implemented in schools.

Present school furniture is often not functionally accessible to children in wheelchairs or even to able-bodied children of varying anthropometric proportions. Available furniture that is adjustable does not accommodate assistive devices, allow wheelchair access, provide easy height and angle adjustability or offer forearm support when using a device such as a keyboard on it. Thus redesign and evaluation of classroom furniture is needed to provide a comfortable work surface that can also be therapeutically supportive.

Video disk and CD ROM technology can potentially be used to store many textbooks, novels and tutorials and present them on command to the pupil. It should be possible to incorporate a space for note taking adjacent to or within the text so that pupils can record their comments or answers to questions. The process of easily retrieving a particular page for display still needs to be determined.

The community environment

The Government of Ontario has been proactive in facilitating wheelchair access to public buildings by funding the addition of ramps, elevators, wider doorways and accessible washrooms. However the corner store and the local bank very often do not cater to the disabled population. Aisles are too narrow for negotiation by a wheelchair. Counters are often too high to permit their use by someone in a wheelchair, and ensure that the wheelchair user is literally looked down upon, and automatic tellers are often out of reach. A more debilitating barrier for disabled persons is that few houses are accessible to frail or disabled persons because of the presence of steep, narrow staircases, narrow doors, and often, small washrooms. As a consequence of this, social interaction between able-bodied and disabled persons is limited to encounters in accessible localities. It is always more difficult and expensive to renovate a house than to incorporate desired features initially. There is a need to develop guide-lines for builders that detail standards to ensure that newly built houses will be accessible by disabled persons.

Mobility

The establishment of the Assistive Devices Programme of the Ontario Ministry of Health to assist in funding selected technical aids has resulted in numerous wheelchair manufacturers seeking eligibility of their products for government support. There is an immediate need for the establishment of assessment guide-lines to permit a review of available wheelchairs to take place. Funding to undertake this should be the responsibility of the respective wheelchair manufacturers or distributors. Aside from mechanical soundness, consideration should be given to the surroundings in which the wheelchair can be used (indoor/outdoor) and whether the wheelchairs can be modified to accommodate growth changes in the user.

A thrust that is presently being pursued by manufacturers of powered wheelchairs is the extension of the driving controls to include environmental control. As previously outlined, it is hoped that the systems being developed will permit a trained therapist or user to modify and customize the characteristics of the control module. It should be possible to operate the controller via single or multiple switches or proportional displacement transducers operated by one or many body sites.

An integral part of a wheelchair that is still in the process of evolving is the seating system. Numerous claims exist about the ability of seating systems to arrest musculoskeletal deformities and position people to maximize their ability to control powered wheelchairs, communication and environmental controllers. Objective assessments of particular seating system features need to be undertaken if the full potential of therapeutic seating is to be realized.

Safety of persons using seating and mobility systems during transport in motor vehicles and public mobility systems is still a concern. Physically disabled infants weighing less than 9 Kg. can usually be accommodated in infant restraint systems by providing simple supportive adaptations which do not compromise the safety function of the restraint. Proven car passengers restraint systems which meet the complex support needs of heavier physically disabled persons are unavailable. Often, straps provided with the postural support systems are depended upon to prevent serious injury or death should an accident occur. It should be possible to develop a restraint system that will interface the postural support system to the car passenger seat and secure the child independently from the seating insert. A similar tie-down system is needed when transporting a child seated in a wheelchair. The tie-down system should safely secure the child without depending upon the structural integrity of the wheelchair.

The Ontario Advisory Councils on Senior Citizens and the Physically Handicapped prepared a report on public transportation in Ontario that recommended, amongst others, the following areas for research and development: improved lift and ramp technology; an efficient, possible retractable, lower step for buses; escalators that are wheelchair and baby stroller accessible; accessible vehicle design; and improved wheelchair construction with respect to safety.

References

1. Symington, D.C., O'Shea, B.J., Batelaan, J., White, D.A., (1980) : "Independence through Environmental Control Systems", Canadian Rehabilitation Council for the Disabled,Toronto.

2. Snell, E., Atkinson, C., Naumann, S., Bugaresti(1987) : "The development and clinical evaluation of a radio frequency linked, computer based, voice controlled workstation for the high level quadriplegic". Final Report for Ministry of Community and Social Services, Applied Program Technology Branch, Toronto.

3. Halpern, C., (1988) : Thesis proposal, Computer Science Department, University of Toronto.

4. Ontario Advisory Council on Senior Citizens and Ontario Advisory Council on the Physically Handicapped (1987) : "The freedom to move is life itself" - a Report on Transportation in Ontario.

General references

1. "The more we do together; adapting the environment for children with disabilities". The Nordic committee on Disability in cooperation with the World Rehabilitation Fund, New York, 1985.

2. "From toys to computers: access for the physically disabled child" C. Wright and M. Nomura, San Jose, CA, 1985.

3. "Aids to make you able: self-help devices and ideas for the disabled", W.M. Davis, General Publishing Co. Ltd., Don Mills, Ontario, 1981.

4. "Living independently", A Shearer, distributed by Oxford University Press, London, 1982.

5. "The accessible home: renovating for your disabled child", H.Snell. Is Five Press, Toronto, 1983.

Reduction of disability - control of the environment

Discussion

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My discussion today is based upon my experience for thirty years working primarily in physical rehabilitation and initially with adults. The last 20 years I have worked with children. I remember the days when either I sawed out wheelchair lapboard from plywood or enlisted the help of the building maintenance person or parent. Things have changed, for out of the consumer need and the availability of new technology and materials, commercially manufactured adapted equipment, mobility aides, environmental control and communication systems are available. The spectrum of current and future devices availability was presented quite thoroughly by Dr. Naumann. With this burgeoning of possibilities, a new profession, rehabilitation engineering, which he represents, has emerged. Such expertise is not always available, even in large medical centers such as the University of North Carolina at Chapel Hill. So, as a practitioner, I must rely on my local durable products vendor to assist in helping to choose items to promote independent functions. Because this is not an uncommon situation, I will address this topic more from a professional consumer's perspective. Suggestions for areas of research will be indicated by problems I have observed from the clinical perspective of a physical therapist.

The title of this session, Reduction of Disability - Control of the Environment, has many implications and nuances of meaning. The individual client is given a means to control his person in space as well as interaction with people and objects in the surrounding environment. Prevention of further disability is also implied. The ultimate goal is for the individual to live a productive, self fulfilling life as independently as possible and in the least restrictive environment. This is true regardless of the individual's age, developmental level, type of physical disability and confounding associated problems. With this in mind, I will begin first with the individual's physical status and the potential for developing deformities.

Prevention of developing deformities is addressed by physical exercise, surgical procedures, general positioning and the use of orthotics. My concern today is orthotics. The primary changes observed in orthotic devices seem an outcome of the availability of new materials. Other than spinal bracing (e.g., thoracic suspension orthoses and the Boston system for idiopathic scoliosis), little has been done in relation to extremity splinting and bracing. The engineering and comfort aspects have not been addressed. Total body contact bracing of the trunk is a problem in the South because of the heat. Children have difficulty tolerating such extensive plastic bracing unless they are kept in an air conditioned environment or unless the bracing is mandatory for post surgical support. Long leg braces and ankle foot orthoses (AFO) using plastic or polypropalene for form fitting do not allow for any significant growth. The cost of any orthotic device at times seems prohibitive especially when it needs replacing every 12-24 months even though the use of AFO's has lessened the need for the very expensive orthopedic shoes of the past.

The use of orthoses is often coupled with the use of a walking aide. Children using crutches can carry books in a backpack if it does not throw them off balance. If so, they may hang a tote bag on the hand grip which often sways and interferes with their walking which is usually tolerable. Students using crutches cannot carry their own lunch trays in the school cafeteria. This can be an issue leading to school Personnel requesting a walker or wheelchair for the child, which, of course, is not always appropriate.

Walking aides is another area in which little has been done regarding design and function other than the recently designed Posture Control walkers by Kaye Products and the posterior Walker by Guardian Products (1) Are new designs of walking aides possible which would free the child's hands and arms for other activities combined with walking at least for short distances?

The use of walking aides does not preclude use of specialized seating and wheelchairs. Thus, when examining environmental controls and interaction capabilities within the environment, I envision it as a fish-swallowing metaphor. The little fish is swimming along and a bigger fish swallows it and still another larger fish swallows that one.

Chairs are another complex aspect of our topic. Is the chair or chairs going to be based on the child's needs and potential for acquiring independent skills or on the caregiver's needs? Hopefully both can be given equal consideration. The child's potential can be overlooked when the house is located in a rural setting and is a small mobile home not accessible to wheelchairs, or when a single parent has a back problem making assistance very difficult or impossible during the learning process of wheelchair transfers. Light weight and small transporting systems consisting of a stroller and seat insert or another type of "transport system" may be selected which does not permit any attempt for independent mobility.

The actual selection of a wheelchair or safety transport chair has become very complicated. The modular designs by various manufacturers permit a mix and match possibility allowing more customized options (2), (3). Not only are there more customizing options available but many more styles of wheelchairs exist, as indicated by a recent spotlight feature in the Journal of Home Health Care and Rehabilitation. Eleven styles of lightweight wheelchairs were featured with descriptions of practical accessories (4).

Perhaps, in some instances, we are using the chair with its scoliosis, head and shoulder support systems as substitutes for the head-to-toe bracing of the past. When we do this, we are immobilizing the child in a sitting position. The child isn't sitting; the chair is holding them in a sitting position much the same as the old bilateral long leg braces with chair back extensions did for standing and sitting. With the braces, a pivot-standing wheelchair transfer to a commode or other furniture was possible. I bring this up not because I want a return to those "old days" but to suggest we have lost a very important possibility in some instances. As physically challenged children grow up, they gain in height and weight and can no longer be lifted in and out of the chairs. Chairs and other devices are needed which promote the transfer of the older child or adult who is dependent upon others to get in and out of the wheelchair and other furniture without undue stress on the assistant's back.

Many seating systems are excellent in providing good body alignment but provide no mobility. This is slowly changing, as standard wheelchair bases are becoming available for use with some of the postural control systems. The awkwardness for the caregiver often remains.

The next level of mobility aides are the variety of powered chairs and the various systems of control. These include controls for operating the chair, household appliances and lighting and interfaces with computer and other technical aides so aptly described by Dr. Naumann.

Now that the chair is selected and all of the needed control systems designed, will the mobile chair fit in the house and in particular in the bathroom, bedroom and kitchen? Are the doorways wide enough? Is the turning space adequate? Can the individual reach objects in the cupboards, dresser, or medicine cabinet. The home now swallows the chair. Pre-training is desirable for the young child who has no sense of balance or righting reactions and does not know that the chair will tip over if not operated properly. This pretraining includes the child's learning to use various types of switches and control devices.

Being socially isolated is a major concern. How is the individual going to get to school or work? Schools now have special buses for the wheelchair bound student but safety in the bus needs to be evaluated. Should the chair be tied down so the child faces sideways, or forward in the same manner as other students? The lift controls need to be enclosed so the young child with exploring hands cannot reach them while in transit.

Public transportation systems have lifts, but are the bus routes available to the disabled? Those who live in rural areas do not, in many instances, have access to public transportation. Families look to special vans which can accommodate the chair. The mini-van is quite popular and is expensive.

The work environment as well as recreation and leisure desires are other considerations. Our ultimate goal for physically challenged children is that they grow-up and enter the world of adult work and community involvement. When assessing all the possibilities of mechanical and technological aides, but concentrating primarily on sitting as a paradigm, we see there are numerous chairs designed for specific environments as well as houseloads and busloads of accessories for environmental control and interaction. Do we need to adopt the Swedish approach whereby each individual has the possibility of getting three powered wheelchairs - one for the home, one for school or work and a third for leisure activities.

In summary, through the use of nuclear age materials, new technologies for various control systems and the emergence of a new profession, rehabilitation engineering, many more viable options are available for the physically challenged child who will grow to adulthood. Researchers need to address some of the more mundane problems related to orthotics such as comfort and allowing for growth, transfer aides for getting in and out of bed or the bathtub, etc. In addition collaborative efforts for developing early training programs for the use of controls and mobility aids are desirable. With these possibilities, especially the early use of powered chairs, care must be taken not to develop a sedentary group of physically challenged adults. Walking is exercise. Aerobic capabilities must be developed in the early years and must be maintained to promote general physical fitness as is recommended for the able-bodied population. Energy studies are needed to establish the energy cost of the use of various mobility aides and at differing ages as well as from a long term perspective of physical fitness. Does the young child who learns to walk as preschooler maintain walking capabilities longer? Are they healthier? Do they learn where and when various mobility aides are most appropriately used? Assessment procedures which include the child's physical abilities, potential problems and the various environments in which he/she must function (home and school) need to be addressed and should include the growth and developmental potential of each child. Are environmental module packages available which would assist the community practitioner in using appropriate technologies for their clients in the classroom, home and leisure activities? Can these technologies benefit the general public much as the curb cut has? This may help reduce the cost and also attract industry to support some of the research efforts. Cost is a major factor when the individual family must finance all the aides which will help their child develop to his or her potential. Some major manufacturing companies, e.g. Apple and Nissan, are looking at the physically disabled population as a profitable potential market. Can these resources be explored to help decrease the cost of the products which enhance the mobility, learning and functional potential of the physically challenged child?

Care must be taken not to overburden the family with aides and gadgets which are not efficient but to select items which will be used. The child and family need to assist and participate in both problem solving and decision making.

References

1. Wilson, J., "Selecting and Using Posture Walkers for Children with Cerebral Palsy," Totline, Vol. 14, No. 2 Summer, 1988, pp 15-16.

2. Norgard, M., "Take a Seat" Rx Home Care, The Journal of Home Health Care and Rehabilitation. Vol. 10, No. 1, January, 1988, p 49.

3. Bergen, A. F., "Putting the Puzzle Together: Using Commercially Available Technology to Create Therapeutic Sitting." Totline, Vol. 14, No. 2, Summer, 1988, pp 10-14.

4. "Spotlight: Lightweight Wheelchairs". Rx Home Care, The Journal of Home Health Care and Rehabilitation, Vol. 10, No.5, May, 1988, pp 68-72.

11. Reduction in disability - advances in technology

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Technology has a significant role to play in reducing the burdens of individual physical disabilities, even to the point of achieving full independence in appropriate circumstances. A recent paper "Critical and emerging issues in rehabilitation engineering," identifies areas of technological endeavour that can be highly supportive of disabled persons.

Stress is laid on the multidisciplinary nature of rehabilitation technology - an important factor in researching further technological developments to facilitate their advances. Rehabilitation engineering services need to be underpinned by appropriate research and development further to assure that modern technology can be best utilised with concomitant economies.

Using the paper as a backdrop, several areas of needed research and support are highlighted in what follows.

Technology Transfer

The paper highlights the need for documentation of clinical achievements with technology and the transfer of technology to the industrial sector so that the availability of relevant rehabilitative products is enhanced. A second paper "A model of a powered upper extremity prosthetics research and development program" reflects a model of research and development that accompanies a local service delivery programme to realize rehabilitative products that can be manufactured locally and marketed world-wide. Given the nature of most rehabilitative products which do not attract a large manufacturing base it can be argued that support should be given to models of a similar kind in specific project areas. Essentially, one endeavours to solve a problem by finding general solutions which then with minimum effort are adapted to individual needs.

Interestingly, the realization of a product from conception to production by external organizations often takes as long as 10 years. Once there is a forged relationship between the developer and the manufacturer the time delays for similar products is likely to be reduced considerably.

Development of researchers

Education and development of researchers to contribute to future advances in rehabilitation technology is of the utmost importance. Recent directions taken by The Easter Seal Research Institute in supporting graduate studentships are to be commended. It is quite evident that with current developments in the Assistive Devices Program in the Province of Ontario, there are substantial needs for relevant personnel not only to appropriately dispense technological devices, but to aid in identifying and developing new technologies.

Explorations in the home environment

An extremely important area for future involvement is in explorations conducted in the domestic and community environs of physically disabled persons to identify relevant technological solutions and products which can be most beneficial to the child and the family. Most often, children and their families are seen by professional staff in the institute or facility where they go to receive services.

Postural support and seating systems

Assessments of ventilatory capability, swallowing and drooling and functional capability need to be made consistent with the designs of seating systems particularly for cerebral palsied children. Often, conflicting requirements are set. For example, bracing an individual for physical support might well compromise breathing. Elucidation of the various issues to yield more pertinent seating designs is important.

The development of seating (postural support) systems amenable to ready and safe transportation in automobiles while meeting relevant provincial and federal safety standards is of growing importance.

Studies relating to the energy levels of disabled persons and strategies to minimize energy consumption are needed.

Augmentative and alternative communication

These areas are taking on greater significance for technology as the promise of computer-based technology is being realized to aid non-speaking physically disabled youngsters. While "Low-tech" solutions should not be ignored there is considerable focus on "high-tech" aspects. Research areas include continuing studies of control methods for access to computers and ways to develop the skills of disabled individuals to best exploit these methods. Of importance also are means for best effecting training of therapists to utilize developed and developing access methods. Explorations with clients are needed regarding acceleration of rates of communication by various techniques including abbreviation expansion or predictive linguistics.

Enhanced use of voice synthesizers to create voice output is also indicated. Investigations relating to voice or utterance control systems also appear to be warranted. Computer-based systems have the potential for educational developments and vocational opportunities. Applied research in these various areas is needed. Aids to facilitate paper and disk handling are needed.

Explorations regarding the social impact and acceptance of use of related technology should be undertaken.

Conclusion

It is hoped that these remarks will be helpful in the promotion of discussion to establish future directions for The Easter Seal Research Institute concerning technology to reduce disability.

References

Mifsud, M., Milner, M., A Model of a Powered Upper Extremity Prosthetics Research and Development Program. I.E.E.E/ Engineering in Medicine and Biology Society Conference, Arlington, Texas. October 1986.

Milner, M., Ph.D., P.Eng., C.C.E. Critical and Emerging Issues in Rehabilitation Engineering. I.E.E.E/ Engineering in Medicine and Biology Society Conference, Arlington, Texas, October 1986.

12. The psychosocial impact of handicap on children and youth

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The psychological aspects of disability might benefit from some brief self-examination. In this regard, Yuker (1988, Attitudes Toward Persons with Disabilities) notes "professional consensus changes over time, but if you take the trouble to analyze the transformations in this practice other than to label the new as innovation or enlightenment we might learn something...few students are interested in developmental accounts of their profession, probably because they do not perceive them as contributing to their clinical skills... most of them (students) seem to assume that what is most recent is most valid and proficient." An analysis of the sciences can provide a particularly promising area in which to explore the role of social forces current in the larger society in shaping the evolution of a discipline. A history of a discipline can tell us important things about where we are going, and maybe where we should be going.

Yuker's current text presents some of Michel Foucault's earlier work. Foucault was a French philosopher and social commentator. His ideas are rather provocative. Foucault presents an interesting view in understanding minority groups, such as disabled. He encourages us to "step back" and examine our daily practices with an anthropological detachment. Foucault notes that as professionals we may oftentimes define the problems, the agenda, and the social reality of those with disabilities or minority differences in ways that serve our own professional interests more closely than those with particular disabilities or low-incidence needs. Foucault noted that "poor people, deviants, and political dissidents" in modern times have been treated with less overt constraints than in earlier times. However, Foucault suggests that, in some respects, social science professionals as well as the technical knowledge may have taken the place of "brute force" in controlling certain people or groups over the more recent decades. In this regard, there is a widening gap between the ideology and culture of professionals and those with disabilities ... they (professionals and those with disabilities) often disagree about "the problem of disability." Science and all disciplines occur with a particular social milieu, and, therefore, the social climate may either enhance or prevent us from seeing or doing what needs to be done in the next twenty years in the area of disability. Lest we not get too far from the point of examining our "power" relationships with those who are less powerful, we need to be reminded that those with disabilities are usually not a self-conscious or well-organized constituency in the same sense as are partitioners, researchers, and academics. Again, as noted by Yuker, "they (those with disabilities) very much depend on the latter (researchers, academics) for changes in the quality of life although there are some notable exceptions. One of the most important features of these minority groups is their autonomy and freedom to address their own needs directly to legislatures and the public, unfiltered by parochial professional interests. For this reason they (those with disabilities) need to attract that minority of professionals who are sensitive to the needs not being met by traditional rehabilitation services, training, research, or funding." Yuker reinforces the notion that the technical and practical knowledge needs of professionals and people with handicaps are often different, a lesson that the larger community of disabled persons maybe has not yet mastered. Lest these comments sound too political, they do reflect a growing awareness among professionals that is needed in inspecting our personal goals as well as the goals of those with disabilities when we begin to plan research agendas.

Clearly the study and the understanding of disability is undergoing some rather significant changes. Several authors (Hahn, 1985; Chesler, & Chesney, 1987) have noted a "definitional shift" from the medical perspective in understanding disability, which focuses only on functional impairments, and an economic approach, which emphasizes only vocational limitations, to socio-political perspective which regards disability as an emerging product of the interaction between the individual and the environment at all levels. This latter view, a minority approach or perspective, has been common in sociology and psychology circles for several years. This type of thinking is well amplified by Yuker's earlier comments as well as some of the cogent writings of Michel Foucault in "The Birth of the Clinic" (1973), as another example. However, this socio-political trend also reflects a resurgence of group identity for those with visible and not so obvious physical differences and in part underscores the basis of the so-called "empowerment movement" (Asch, 1984). Here, individuals are encouraged to "take charge of their lives" and become more part of the treatment establishment, that is, to empower themselves. Policy makers are beginning to recognize these impacts. Witness the recent concern at Caudellet in the United States by the hearing impaired students for a President more sensitive to their needs. Although this trend is likely multifaceted in its origin, it does continue to emphasize that those with handicaps live with a particular stigma, a noticeable difference, which is molded and shaped by the attitudes of others within a social-interpersonal context. These aforementioned issues seem to imply that there are some new trends, or at the very least, a different focus to possibly consider in the area of research as well as the treatment partnership for rehabilitation for those with disabilities. On a practical level, we need to pay much more attention to the relationship between training, practice, and reality. Yuker notes "the current difficulty of rehabilitation emphases is not an ephemeral phenomena generated only by a business cycle or by rapid technological change, or even by the new conservatism in North America. Rather, the impasse is rooted in the structure of vocational rehabilitation services as defined in the state-federal programs, and in the kind of graduate training often mandated for professionals entering those programs." There is often a discontinuity between existing training models and what is occurring for those with a disability on a daily basis.

The focus

Efforts in the Reduction of Handicap should logically consider the social and psychological consequences of these "differences of person" as they exist in the person to person environment at all levels, social and political. More specifically, attitudes of able-bodied towards those who are less able and self-attitudes of those who are disabled can have a major and evolving impact on the definition, adjustment and meaning of the term "handicapped". In particular, social and legal changes have resulted in a modest integration for those with disabilities, assimilation is promoting some better adaptation, however the stigma of disability continues and will likely always be an issue. Reduction in Handicap should consider the psychosocial implications of disabilities as part of the management and remediation process. This paper will attempt to examine some of these key issues in the study of attitudes of children and youth with disabilities and suggest directions for future research and thinking. Professionals in the area of disability research need to periodically do a historical autopsy of their discipline and profession. Science and empirical research can become dogma. We need to be clear that we are not simply perpetuating or just reflecting societal difficulties by discursively clarifying them, but hopefully adopt an attitude of professional activism to more directly clarify the issues of disabilities and in concert with practical demands of those with disabilities.

A few organizing principles should be mentioned briefly at the outset. Much of our applied social-psychological studies of different disabilities over the last forty years has been directed at understanding the determinants and dynamics of stigmatization; the study of prejudice. The vast majority of these studies have repeatedly identified the negative consequences of being different. This problem (prejudice/stereotypical attitudes) is likely an inevitable consequence or feature of social groups. Some, possibly many professionals and lay persons, view this fundamental negative bias (Yuker, 1988) as an ingrained social imperative. This fatalism often appears to surface as one of the major reasons why attitudinal and psychosocial studies are not pursued or broadly supported at governmental levels. furthermore, many of the problems with disability directed social-attitude research can be in large part attributed to the rather pervasive tendency to ask the wrong question. Many studies are not "crisp" in their outcomes or practical in their recommendations. Both of these critiques are valid but tend to deny the importance of such studies and their implications for social policy in the lives of those with differences from the norm.

In the seminal work Social Stigma: The Psychology of Marked Relationship, Jones et al. (1984) and others raise some very fundamental questions: Is destigmatization an important social objective? Are the conditions for minimizing stigma worthy foci for research and social policy? Jones, et al (1984) responds with three cogent reasons to these questions: 1) Negative and erroneous interpersonal attitudes toward those who are "different" perpetuate social distancing between all who do not agree with us; 2) Group - that its, local group and national group - solidarity can be promoted by enhancing attributional clarity and understanding of the acceptance of the values of diversity: and, finally, 3) we need to reduce stigma of difference because it can reduce the psychic pain of feeling rejected and discredited, These arguments underscore how such studies can enhance the "quality of life" and reduce the impact of handicap. They have implications for all members of society.

These prior comments are focused on emphasizing the importance of examining our own inherent biases in doing research "on the disabled," our need to explore such biases, and be aware of how we may perpetuate myth. Secondly, new trends are emerging in the social psychology of disability. Empowerment is occurring for those with disabilities by their accumulation of knowledge and acculturation of governmental systems that are beginning to focus more clearly on the needs of those with particular problems. Finally, it is relevant to study the socio-psychological implications of disability as a means to obtain a more clear understanding of the relationship between those who are able and those who are less able and to promote more effective management in rehabilitation.

Two sections of review of research issues follow: the first section outlines information in terms of "what we know." This section includes some of the more "robust" findings from the literature, those that have some generalization power; however, this is not necessarily an exhaustive listing. Following this section, a review of the problems and issues in the area of research on attitudes toward children/youth who are disabled is presented. This paper concludes with a series of future directions for research.

What do we know?

"Thirty-five years of research on attitudes of others (adults and children) toward those with disabilities (all types) has indicated a general negative bias toward those with disabilities," Barker, R.C., Wright, B.A., Meyerson, L, and Gonick, M.R, (1953). "These negative attitudes are noted to be related to sociocultural and psychological factors, conscious and unconscious issues, experiential and unique factors of the non-disabled toward the disabled," (Livneh, 1985), A basic issue in the psychology of disability relates to our understanding of what Yuker has labeled, "The Fundamental Negative Bias." This attitude has an insidious relationship to much of our thinking, our training, and oftentimes, shapes the directives of our research. As noted by Yuker, "the basic proposition of the fundamental negative bias states that if, for whatever reason, something is perceived as negative (value aspect), and stands out (saliency aspect) in a sparse context (internal or external), then the psychological events that follow will assume a negative course. In effect, the resulting attitudes are negative." Similar concepts have been reported by several writers in the disability literature and described as the negative spread effect of a particular type of disability or disabling condition on the person with a disability: the disability invades the "personhood." The importance of this concept is that it not only exists as a social psychological phenomena, it also exists as a latent characteristic guiding our research and our thinking about people who are different.

Physical appearance, which includes voice, action, movement, etc., has been documented as having a profound impact on others' social and evaluative reactions, others' ratings, interpersonal acceptance, and the mental health adjustment of those with visible disabilities. The majority of this "attraction" research began with the work of Dion, Berscheid and Walster in 1972 and was extended to those with disabilities by Richman and Harper (1979), among others.

Methodological cautions about attitudinal studies and disability findings; the following represent some of the more noteworthy cautions in disability attitudinal research, (l) The majority of studies in the social psychological disability literature are correlational in nature with relationships identified between particular variables and characteristics. It must be repeatedly emphasized that significant or high correlations or associations do not imply causation. This is a fundamental problem in over-interpretation of research generally. (2) Attitude tests, when constructed for the study of disability, should have an equal sampling of both positive and negative references or questions to the disability or the disability in question. Without such a balance, the fundamental negative bias may operate and confound the results. (3) Much of attitude disability research is characterized by studying labels as categories of various groups. Researchers are cautioned against the tendency to interpret the findings about a particular disability in terms of the characteristic associated with or indicated only by the label. Such research perpetuates the "label mentality" (i.e., retarded are __________, disabled are ___________, etc.), (4) Finally, and rather importantly, significance levels (alpha levels, .0l, .05) should not be equated with the robustness or importance of the study findings. Put somewhat more directly, the strength or size of the level of significance is not the same as the level of practical significance for a particular finding.

Disabilities that reduce mobility oftentimes promote a homogeneity of social isolation. Such individuals are often rated higher on measures of internalising their difficulties. (Harper, 1978, Richman & Harper, 1980).

Specific disabilities are generally unrelated to particular personality types. This has been well-documented for the last fifty years. It is more important to examine the fact of disability than the type of disability (Harper, 1978, 1980) when assessing adjustment correlates.

Mental health difficulties, psychosocial stress, adjustment problems are associated with individuals who have a physical disability and also at a higher rate. The risk of emotional dysfunction for those with such chronic illnesses and physical disabilities ranges between two and five times greater than their healthy peers, depending on the functional impact of the existing disorder Cadman, Boyle, Szatmari and Offord (1987). Cadman, Boyle, and Offord (in press) have also explored mental health risks for siblings of handicapped. Additional support for this risk factor being associated with physical impairment is also noted in the work of Pless (1971). Anticipatory guidance does have benefits for those with physical and visible congenital impairments. Specifically, there appear to be predictable stress points occurring within a developmental framework (predictable transition times) for children and youth. Consequently, anticipatory review, guidance, parental and child preparation for such transition points appears to have support and should be routine practice (Wright, 1980).

As noted by Yuker (1988), "the attitudinal consequences of contact with disabled persons are mediated by characteristics of the disabled person and the interaction." Data for contact to result in positive attitudes for the disabled individual implies that the disabled person should be perceived as competent in the areas that are valued by the other parties in the interaction, socially adept in communication, and open to the discussion (be able to describe) of their particular disability. Disability characteristics appear to be most salient in affecting initial contacts. However, as contact is extended, particular aspects of the disability become less salient and interfering in social interactions. The non-disabled person will likely develop positive attitudes toward disability if 1) the disability is not the most important characteristic in the interaction, 2) a recognition exists of the heterogeneity of disabled people, and 3) there is an equality of status in the relationship. For social interactions to promote positive attitudes, they should be cooperative and reciprocal, mutually rewarding and have some stability of purpose. Overly contrived contacts have limited benefits generally. Implicit in this listing of characteristics which promote and clarify contact with the disabled is that contact under specific conditions can and does have a positive effect on promoting positive relationships between those who are disabled and those who are non-disabled.

A considerable amount of research regarding elementary age children's attitudes toward disability is presented in the work of Steven Richardson (1976). Richardson used a series of line drawings of common visible disabilities, and presented these to disabled and nondisabled children to learn about their reactions to such differences. Consistent findings across age, cultural, and disability groups led Richardson to conclude that both disabled and nondisabled children prefer nondisabled peers, that there is considerable agreement among children as to their preferences for particular types of disabilities over others, and that different disabilities very likely connote different degrees of stigma. This research has been extended and clarified, but it is predominantly analog (pictures of situations) in nature. Such picture ranking tasks assume a reality and connection between expressed verbal preference and actual daily behaviour. Nevertheless, the findings of Richardson are potent and generalizable, emphasizing a hierarchy of attitudes and preferences among children who are disabled and nondisabled. Cobb and Hancock (1984) further note that the evidence of social isolation of disabled children is supported by research with children and teenagers using many attitude scales. These studies have been conducted viewing attitudes toward the disabled on uni-dimensional continuum, positive to negative. Results suggest that as the child progresses to the teenage years, attitudes toward those with visible (noticeable) differences become more rigid, negative, and rejecting. The teenage years appear to be the most difficult time for adjustment for those with any type of disability. It is clear that even at early ages, children's responses to the disabled are frequently negative and rejecting or, at the very least, ambivalent. Children seem to be able to and do make distinctions on the basis of physical attributes at an early age, and express preferences for nondisabled peers, as a result.

Kagan et al (1966), in a series of studies with human infants, determined that very early in life we may develop a "set of perceptual expectations" of what is characteristic or pleasing of a human face. Consequently, responses to disfigurement differences, may have a certain perceptual (inborn) basis, or at the very least, are present at a very young age. Much of Kagan's research is with infants under four months of age. More recent research has extended this to younger, presumably naive infants and their ability to recognize and/or respond to dysmorphic drawings of faces, suggesting a perceptual "set".

N.J. Lerner (1970) began a series of studies which continued for the next decade examining many prevailing assumptions that the world is a benign or at least a just place. The importance of this concept, which came to be known as the "Just World Belief" relates to our social understanding of why we believe negative things seem to happen to certain individuals and not others. This social psychological concept is useful in understanding how nondisabled individuals interpret the characteristics and behaviour of those who are disabled, different, or not part of the "in group". In brief, individuals are motivated to believe that the world is a stable and orderly place and that there is a general approximation between a person's character or actions and their outcomes. (This belief is not usually held in Eastern cultural groups). This belief is important since it permits us to believe that our own actions and character will be rewarded and that we have some control over our lives. Lerner has proposed that the existence of this need predisposes people to hold unjustifiably negative evaluations of victims of misfortune or injustice. Beliefs that members of the victim groups are weak, unworthy, foolish, careless, or otherwise different allows us to distance ourselves from those with the differences (all types) and assists us in believing that we can avoid a similar fate. It is relatively complicated concept with many implications, it has been researched with a variety of populations, and is assistive primarily in clarifying some of the underlying assumptions of the nondisabled toward the disabled.

There is a significant social distortion message potentially present in the interaction between an individual with visible (or somehow noticeable) physical impairment and those who are nondisabled. This message has often been referred to as noncontingent feedback. The distortion of information given the handicapped person by the nonhandicapped person, because he does not wish to offend or hurt the handicapped person, may result in the handicapped person not obtaining honest or situationally related-feedback when he/she behaves inappropriately or appropriately in a particular situation. This reduces the likelihood of learning appropriate and inappropriate behaviour within certain social contextual situations. This is an extremely important concept to understand the behaviour of those who have visible or noticeable differences (Richardson, 1976). There are many corollaries and related issues to this phenomena that can reduce social contact, promote withdrawal, decrease self-esteem, and generally distort the interpersonal feedback between someone with a visible or noticeable difference and those who are not disabled.

Richman and Harper (1979) noted that individuals with disabilities often experience difficulty in incorporating their stigmatizing conditions into their overall self-view. Clarifying this idea a bit, as we develop our self-esteem and self-concepts, we define ourselves around "what I am" and "what I can do." In this regard, we form definitions about ourselves on the basis of what we are. We usually do not form clarifications about ourselves in terms of "what we are not." If, however, a negative aspect of self does become important in one's own and others' perceptions, it may be necessary to create some type of structure around it: and the question for the person with a disability remains, what is the nature of this structure of self-knowledge? This theorizing is outlined by Jones, et al., in his 1984 text. He notes, "individuals who are fat or crippled or deaf may (be socially forced) to think in terms of what they are not or what they cannot do, such as 'I am not thin, I cannot wear nice clothes, I cannot dance, I cannot be accepted'. This type of self-knowledge explicitly invokes contrast with other people. It may be that a personality structure based on such negative self-knowledge is less stable than one based on positive self-knowledge, and more directly tied to social comparisons with past selves." The bottom line is that it is extremely difficult to construct a concept around a void. However, this appears to be what many individuals with significant impairments may actually do in part psychologically. Jones et al. (1984) also asserts that certainly disabled individuals do build self-concepts around positive traits. However, there is often an asymmetry of assumptions between those who are disabled and those who are nondisabled conceptually in terms of interpersonal expectations. That is, the nondisabled focuses on the potentially stigmatized individual and sees what the individual does not have or is not capable of doing. This attitude often dominates the nondisabled person's conception of those who are disabled. For the disabled, however, the focus is more likely to be on new and different abilities (change and progress) that represent an adaptation to this earlier view of difference. This potential asymmetry between the disabled and the nondisabled in what each assumes about disability and what is central to the self-conception is one of the stumbling blocks to relationships between those who are disabled and those who are not. That is, it is difficult for each to grasp the assumptions of the other.

Evidence is accumulating that participation in athletics for those with disabilities appears to be enhancing in all respects. Specifically, athletes at all levels of disability appear to report more satisfaction with life, more general happiness, and generally appear to develop more assertive social and living skills (Valliant, et al., 1985; Emes, 1981).

As noted by Hannah (1988), teachers can have an extremely potent effect on the attitude development of children with all types of disabilities. Hannah's review of research indicates that teachers can be generally negative about beliefs and feelings toward the children with disabilities and presents evidence to support a general reluctance of teachers to enter into more "intense" teaching relationships with those who are disabled. Teachers represent another group with generally similar public attitudes toward those who are disabled. Probably the most significant consequence of teacher attitudes toward disabled children has to do with expectations and ultimate levels of achievements both cognitively as well as psychosocially. A current review of the literature indicates that teacher knowledge of disability and its implications remain among one of the most significant stumbling blocks to promoting independence in children with all types of disabilities. Peer to peer interactions hold considerable promise for improving the attitudes toward elementary and junior high aged children. Rosenbaum and colleagues (1986, 1987) rather convincingly demonstrate that a "Buddy Program" (randomly selected buddies paired with a particular gender-matched disabled child for a three month period) had significant and long-term (Personal communication, 1988) effects on improving attitudes of the nondisabled consenting volunteer. Such potent classroom-based experiences hold considerable merit for future programmatic implications of improving attitudes within our public educational systems.

Summary of what we know

There is a fundamental negative bias that appears to operate in all aspects of dealing with disability (all types).

Physical appearance is a very significant factor in all aspects of disability adjustment.

Particular disabilities do not lead to disability-specific personality types.

Individuals with handicaps (physical-all types) are at a higher risk for mental health adjustment concerns.

Anticipatory guidance has benefits for those with physical impairments.

Certain types of peer to peer contact with individuals with disabilities does promote more positive evaluation of those with impairments.

Children's preferences have been established for particular disabilities. Different disabilities seem to reflect a hierarchy of acceptance by nondisabled youth.

There may be a cognitive perceptual set, as well as a social basis for our reactions to those who appear different.

People with physical differences are often devalued as "defense" by those who are not disabled.

The interpersonal communication between the able-bodied and the disabled is often based on "disinformation."

Positive self-constructs (what am I) are more difficult to formulate for those with physical impairments.

Teacher knowledge of disability can have a significant impact on disability adjustment.

What we do not know

(A brief review of the problems and issues in the area of research and attitudes and behaviour toward children and youth with disbilities)

As noted in the majority of research (fact of disability) reported previously, much attention has been devoted to the label of disability or various disability labels (type of disability). Much preoccupation exists with the initial consequences of labels on behaviour. However, little attention has been given to the stimuli used to elicit these attitudes or that seem to underlie these particular labels. Richardson (1976) has maintained that for some time research has focused too much on the verbal label to the exclusion of the handicap and everything else about the person in trying to study the issues. Research efforts now need to move beyond this fairly simple-minded correlational labeling of groups in an attempt to look for significant findings which try to change these labels and assume that changing a label changes these other social/interpersonal attitudes.

The majority of handicap disability research is also analog in nature. That is, it attempts to assess attitudes through some survey, interview technique, pictures, or it uses confederate (fake) disabled individuals. It is often assumed that there is a relationship between verbal report in the experimental situation and actual behaviour (e.g., "What you say is related to what you do."). This is probably the most difficult social-psychology assumption and the most difficult prediction question in our study of attitudinal research. Future research needs to emphasize the social context or the setting in which attitudes or behaviours are being displayed toward those who are different and adopt a more comprehensive review of disability beyond the label to include movement, gesture, speech, and other aspects of behaviour. Richardson (Personal communication) is exploring a comprehensive disability index that includes over 20 different aspects of the person and the specific environment. Observational studies are also needed in this area that assess on-going "live" relationships between disabled and nondisabled children we need more randomized controlled treatment studies examining attitudes, behaviours of children in classroom, play, and living settings. These "in vivo" studies are currently best represented by Rosenbaum and his colleagues.

As noted by Yuker (1988), Jones et al. (1984), and Richardson (1976), the majority of experiments in this area have a common tendency to get preoccupied with the presence or absence of handicap and pay little attention to the characteristics of the disabled individual, the nature of the interpersonal relationship, or the setting in which the social encounter occurs when studying disability issues. Harper, Wacker, and Cobb (1985) note that changing the social situation affects the nondisabled child's preference or acceptance of a particular disability. These social interactions are very complicated interactions that are rarely well presented in research proposals. Research in this area needs to more adequately assess and include the social and physical contexts of encounters between disabled and nondisabled if we are to get a better understanding of what is going on in peer to peer contacts. In this respect, we need to look for compensatory characteristics that may reduce the saliency of a handicap in particular social or interpersonal situation. It is no longer relevant to identify that it is socially disadvantageous to be physically or psychologically different, and we must move beyond this conclusion.

A final methodological area in reviewing this earlier research relates to the complete lack of studies on children with disability as they have grown up through adolescence and adulthood. We seem to focus on very convenient samples, yet it must be recognized that it is very difficult to get a coordinated prospective longitudinal study going since most funding agencies are not currently interested in this type of long-term commitment. We sorely need studies on adults and elders as they age and deal with disability within the aging process.

Future research - where do we go from here?

There is a major discontinuity between what is taught in rehabilitation training programs and what is needed in practice by the predominant and increasing group - the more severely disabled. Graduate training (all types) is still focused on those with marginal or lesser disabilities. This is true at all levels of education. Research needs to be directed at exploring more innovative "model curriculums" and providing experiences (exposure to disabilities during training) for professionals that are well-known to shape later professional attitudes towards those with disabilities.

Research proposals on attitudes and treatments toward disability should also include more complex social situations. That is, we must move beyond label identification and attempt to isolate those factors that people are responding to in social situations that promote more positive social contact.

A number of studies have indicated that certain moderator variables (social skill training, appearance management, sibling support, informal family support networks) tend to offset the negative effects of growing up with physical impairments. These studies that outline the more positive strategies for dealing with differences are often referred to as "coping studies." Such positive strategies also include training for those with disabilities in clarifying (telling about my disability) the particular disability, behaving in a positively assertive fashion, and demonstrating social competence. Many more studies (live interactions between those with disabilities and nondisabled) are needed that examine these specific kinds of competencies and behaviours that promote teaching of these types of positive coping strategies to individuals with physical differences.

The effect of handicap on behaviour has been studied under conditions where other factors (age, sex, type of disability) which influence interpersonal behaviour have been held constant. It would now be valuable to begin to systematically vary other factors to determine the extent to which changes in behaviour toward the handicapped are associated with other characteristics of the handicapped person and the social and physical context in which the social interaction occurs. As an example, there is evidence that interpersonal behaviour is clearly affected by how "good looking or attractive" one person appears to another. However, attractiveness is also related to the self-perceived level of attractiveness of the rater as well. Appearance management has become a favourite term for individuals interested in promoting positive aspects of how one looks in a social situation. A more complex design could examine combinations of different characteristics of a person, varying characteristics such as cues of movement, role, and nature of the interpersonal task, as well as intellectual abilities as displayed, skin colour, etc. The major point here is to use a more complicated design to sort out these more complicated questions. Video presentations are useful for research and training purposes.

Very little effort has been directed in the education of children (nondisabled) to teach understanding or appreciation of individual differences, to discuss questions dealing with differences in race, intelligence, wealth, physical handicap, or behavioral disturbance. My own clinical observations as well as the research of Richardson tends to indicate that children in the elementary grades are eager to discuss these types of interpersonal questions. We need to explore how children gain an understanding and a concern for individual differences and how one might better prepare them for more satisfactory and sensitive interpersonal relationships where the more salient issues of interaction are rarely addressed. Quite basically, we need to learn more about the sources from which children derive their early learning about physical handicaps and how to help parents of those with disabilities deal with normal questions from their children that are raised in peer-to-peer contact.

At another and earlier level, we need to explore what forms of discussion (guidance) with parents of handicapped infants can be helpful. Although we have evidence to suggest that anticipatory guidance and shared decisions around treatment is useful, we are unclear on what particular issues to emphasize for parents of young handicapped infants, often times parents maintain a certain attitudinal set based upon their ideas of a premature or difficult birth or an early label (mental retardation, brain damage), which often interferes with their natural developmental aspects of parenting. That is, because of a difficult birth and parental perceptions surrounding these difficulties, the parents themselves may adopt a somewhat stereotypical response which can alter their expectations and caring behaviour for their infant. In this regard, Jensen, Harper, Lindgren, Blackman, and Hein (1988) have identified a "Prematurity Stereotype" that appears to be operating in the care giving of parents with "high-risk" infants. Some of the more interesting research that has developed recently in this area also relates to the problems of "depression" and lack of affective responsiveness of young severely physically impaired, but not mentally retarded, handicapped infants. Because these infants are generally viewed as passive, parents often do not interact with them and frequently tend to withdraw from them. These infants may develop "depressive-like" responses. We need to clarify this relationship much more specifically for parents and be more clear in our guidance for these parents with severely handicapped infants. We need controlled studies exploring more specific cognitive and experimental stimulation for severely handicapped infants.

There is also evidence to suggest the handicapped child needs a higher level of social competence to overcome initial barriers created by the particular handicap (Richardson, 1976). Because of the noncontingent and often inappropriate social feedback that many handicapped children receive, social experiences necessary for developing pro-social competence is often lacking or confused, at best. We need to learn what are considered desirable personal characteristics among children and how these may vary in subcultures within our own society. In essence, we need to explore more beneficial ways of educating for tolerance.

As noted by Jones et al. (1984), if we wish to promote the destigmatization, we will likely have greater success by placing those with disabilities in more advantageous situations than by "preaching tolerance for diversity". The television medium, in particular, seems to offer unlimited opportunities for presenting those with various disabilities to a larger number of people. The vivid prototypes provided by such television portrayals can exert their influence rather dramatically. Those of us at Iowa draw on the concrete, vividly remembered case of "Bill" (portrayed by Mickey Rooney). Is there any good evidence that television really makes a difference in this destigmatization process? The existing evidence shows that television clearly has an impact, but it is not always in the direction of increased tolerance. It is, however, a very potent medium. Clearly, more research needs to be directed to this medium, both in educational circles within the schools, but as well on a wider basis for those who have influence in this policy arena.

Research needs to be directed to more studies clarifying how to maintain long-term sibling relationships among those who have disabilities in families. We need to understand how informal support networks of families can be a major positive coping factor and maintained over the years in promoting the more positive adjustment on a continuing basis for those with handicaps. In essence, we need to develop studies that identify positive characteristics of families and enhance such characteristics or at least clarify them so that we know what they are once we see them. Research that identifies how agencies can promote and maintain family involvement in treatment placement is needed. How can we prepare families for transitions, what are the ways to encourage joint decision making, and how can we keep siblings from withdrawing or "burning out" from the severely handicapped brother or sister?

A largely forgotten group in this area of psychosocial research are those who are aging with various physical handicaps. we are now becoming aware of the increasing numbers of individuals with mental and physical handicaps that are surviving into their seventh and eighth decades with very significant handicaps existing from birth. This has not been true in earlier times, and it requires a rethinking of our services, our treatment, and our longer term commitments within our general institutions and support systems, whether they be families or agencies. Such longevity and increasing numbers raises potential concerns for competition of funds and now how to best integrate those who are aging with handicaps into the general aging social systems.

Finally, research programs for those with physical impairments need to be based in a much broader programmatic perspective. That is, agencies need to develop agendas for the future that are based on long range projections, and are based on the needs of an ever evolving constituency. This constituency must be involved directly in this process. This is not an easy task; however, psychosocial research in this field cannot be simply left to the vagaries of individual researchers based on their own creative focus oftentimes fostering their particular "pet" ideas. We need to begin planning more coordinated agendas which involve planners and participants at many different levels and with a variety of disabilities and needs.

References

Armstrong, R.W., Rosenbaum, P.L., & King, S.M. (1987). A randomized controlled trial of a buddy program to improve children's attitudes toward the disabled. Davelopmental Medicine and Child Neurology, 29, 327-336.

Asch, A., (1984). The experience of disability., American Psychologiest 39, 529-536.

Barker, R.G., Wright, B.A., Meyerson, L., & Gonick, M.R., (1953). Adjustment to physical handicap and illness: A survey of social psychology of physique and disability (Revised edition) New York; Social Sciences Research Council.

Cadman, D., Boyle, M., Szatmsri, P., & Offord, D.R., (1987) Chronic illness, disability, and social well-being; Finding of the Ontario child health study, Pediatrics, (79(5), 805-812).

Cadman, D., Boyle, M., & Offord, D.R, (In press) The Ontario child health study: Social adjustment and mental health of siblings of children with chronic health problems. JDBP

Cobb, L.S., & Hancock, K.A., (1984). Development of the child with a physical disability. Advances in Developmental and Behavioral Pediatrics.5, 75-107.

Dion, K.K., Berscheid, E., & Walser, E., (1972) What is beautiful is good. Journal of Personality and Society Psychology.24, 285-290.

Donaldson, J., (1980). Changing attitudes toward handicapped peers: A review and analysis of research. Exceptional Children, 46, 504-514.

Emes, G., (1981). Fitness and the physically disabled: a review. Canadian Journal of Applied Sports Science, 6, 176-178.

Foucault, M., (1973). The birth of the clinic: An archeology of Medical perception. (Translated from the French by A.M. Sheridan-Smith). New York: Pantheon Books.

Harper, D.C., (1983). Personality correlates and degree of impairment in male adolescents with progressive and nonprogressive physical disorders. Journal of Clinical Psychology 39(6), 359-867.

Harper, D.C., & Richman, L.C., (1978) Personality characteristics of physically impaired adolescents. Journal of Clinical Psychology, 34, 336-342.

Harper, D.C., Richman, L.C., & Snider, B.,(1980), School adjustment and degree of physical impairment. Journal of Pediatric Psychology, 36(3), 668-671

Harper, D.C., Wacker, D.P., & Cobb, L., (1985). Children's attitudes toward disabled peers and the effects of mainstreaming, Academic Psychology, 7, 87-98.

Harper, D.C., with the assistance of D. Wacker & L, Seaberg-Cobb, (1986). Children's social preferences toward peers with visible physical differences. Journal of Pediatric Psychology, 11(3), 323-342.

Jensen, A., Harper, D., Lindgren, S. , Blackman, J., & Hein, H., (1988). Prematurity stereotype: Fact or fiction. Paper submitted to the American Academy for Cerebral Palsy and Developmental Medicine, Toronto, Canada.

Jones, E., Farina, A., Hastdorf, A., Markus, H., Miller, D., & Scott, R., (1984). Social Stigma: The psychology of marked relationships New York: W.H. Freeman and Company.

Kagan, J., Henker, B.A., Hen-Tov, A., Levine, J., & Lewis, M., (1966). Infants' differential reactions to familiar and distorted faces. Child Development, 37(3), 518-532.

Lerner, M.J., (1970). The desire for justice and reactions to victims. In J. Macauley, and L. Berkowitz, Altruism in helping behavior, New York: Academic.

Livneh, H., (1985). Death attitudes and the relationship to perceptions of physically disabled persons. Journal of Rehabilitation 51, 33-41.

Pless, I.B., & Roghmann, K.J., (1971). Chronic illness and its consequences; Observations based on three epidemiologic surveys. The Journal of Pediatrics 79(3), 351-359.

Richman, L.C., & Harper, D.C., (1978). Observable stigmata and maternal behavior, Cleft Palate Journal 15, 215-219.

Richman, L.C., & Harper, D.C., (1978). School adjustment of children with observable disabilities. Journal of Abnormal Child Psychology, 6(1), 11-18.

Richman, L.C., & Harper, D.C., (1979). Self identified personality patterns among children with varying degrees of facial or orthopedic disfigurement. Cleft Palate Journal 16(4), 257-261

Richman, L.C., & Harper, D.C., (1980). Personality profiles of physically impaired young adults. Journal of Clinical Phsychology, 36(3), 668-671.

Rosenbaum, P.L., Armstrong, R.W., & King, S.M., (1986). Improving attitudes toward the disabled: A randomized controlled trial of direct contact versus kids-on the block. Journal of Developmental and Behavioral Pediatrics, 7(5), 302-307.

Sheridan, A., (1980). Michel Foucault: The will to truth. London: Tavistock Publications.

Wright, B.A., (1983). Physical disability: A psychosocial approach (second edition). New York: Harper & Row.

Selected readings in attitudes towards disability

Annotated Bibliography

Armstrong, R,W., Rosenbaum, P.L., & King, S.M. (1987). A randomized controlled trial of a buddy program to improve children's attitudes toward the disabled. **Developmental Medicine and Child Neurology**, 29, 327-336.

The study involved random assignment of consenting children in five grades to buddy (with children with a disability) or control groups. The focus of the intervention was with children with the lowest initial expressed attitude ss measured by the authors using their attitudinal measure. The results of the study provided evidence that a direct contact buddy system significantly improved expressed attitudes. This represents one of the few randomized trials in attitude measurement intervention in the literature. The authors note that success in the school environment may be critical to the disabled child's development.

interventions such as the buddy system may increase the chance for establishing successful social relationships with able-bodied peers.

Boyle, M., Tebbi, C.K., Mindell, E.R., & Mettlin, C.J., (1982). Adolescent adjustment to amputation. **Medical and Pediatric Oncology**, 10, 301-312.

Twenty-seven adolescent patients with limb amputations due to cancer were compared with a control group of eight patients who had lost limbs due to trauma. The average age for cancer-related amputees was 23 years at the time of interview, and 19 years for the trauma-related amputees. The mean age at surgery for both these groups was 15 years. The primary goal of this study was to investigate the psychological effects of the amputation procedure on these patients. Factors considered between groups were marital status and family related issues, education, occupation, and employment. Issues of primary concern for the amputee were prosthesis and phantom pain, as well as pre-operative and post-operative procedures. Thoughts about the future and basic adjustment to this situation were also discussed. Results indicate that cancer patients generally had a positive attitude about life, and 85% of this study group were found to have what was assessed by the interviewers as adequate overall adjustment. The cancer patients, as opposed to those in the control group, evidenced a better adaptation to disability.

Breslau, N. (1985). Psychiatric disorder in children with physical disabilities.**Journal of the American Academy of Child Psychiatry**. 24, 87-94.

Psychiatric disturbance in children with four congenital conditions was compared with that of physically unimpaired children and across conditions, three that involved the brain and one that did not. The role of mental retardation and severity of handicap with psychiatric disorders was examined. The pattern of disorder of psychiatrically impaired children with a disability was also compared to their counterparts who are non-disabled. Subjects included families of patients 3-18 years of age. A control group was established, which was comprised of a representative sample of all families in the Cleveland area with one or more children 3-18 years old. Psychological functioning was measured by the Psychiatric Screening Inventory (Langner et al, 1976). Results indicated that children with chronic disabilities were at increased risk for severe psychiatric impairment. Severe psychiatric impairment was higher in conditions involving brain abnormality. Also, severely retarded children were twice as likely as children with no mental retardation to be labeled severely disturbed. However, the severity of the physical disability had little effect on psychopathology.

Breslau, N., Weitzman, M., & Messenger, K. (1982), Psychologic functioning of siblings of disabled children. In S. Chess and Thomas (Eds.), (1982), **Annual Progress in Child Psychiatry and Development**. New York: Brunner/Mazel.

The purpose of this study was to report on the psychological adjustment of normal siblings of disabled children. Results were compared to data on 1034 randomly selected children from a cross section of Manhattan households. The study groups consisted of families of children with cystic fibrosis, cerebral palsy, myelodysplasia, or multiple handicaps. All had been patients at two of three tertiary hospitals in the Cleveland area. Siblings' psychological adjustment was measured by the Psychiatric Screening Inventory (Longner et al, 1976). Results indicated that siblings of disabled children did not manifest higher rates of severe psychologic impairments or greater overall symptomatology when compared to the control group. Siblings of children with disabilities did score significantly higher on scales measuring interpersonal aggression with their peers and within the school. The type of the child's disability was found to have no relationship to the psychological functioning of the sibling. Birth order and sex of the sibling were significantly interactive.

Bryan, D.P., & Herjanic, B. (1980). Depression and suicide among adolescents and young adults with selective handicapping conditions. **Exceptional Education Quarterly**, (2), 57-65.

This article discusses the relationship between various types of handicaps in adolescents and young adults, and depression suicide attempts, and suicide acts. Factors such as physical health, life satisfaction, and adolescent behaviour in the handicapped are considered. The authors present a literature review of this material. An extensive list of behaviours or symptoms that may assist practitioners is given. Authors note that, although handicapped adolescents face unique developmental problems that vary with the nature of their handicaps, the association between handicaps and depression suicide attempts, and suicide has not been clearly established. The paucity of systematic, controlled research on this topic, and the need for expanded research is noted.

Grand, S.A., Bernier, J.E., & Strohmer, D,C. (1982). Attitudes toward disabled persons as a function of social context and specific disability. **Rehabilitation Psychology** 27(3), 165-174.

The main purpose of this study was to examine the nondisabled person's attitudes toward the disabled, across different social situations and with different disability groups.

In order to test the Hypotheses concerning social situations and disability types, an instrument was constructed by the authors. Doctoral level psychologists participated in generating 31 items for this protocol titled the Disability Social Relationships (DSR) Scale. Social situation subscales of Work, Dating, and Marriage were included. Disability subscales of Amputee, Epilepsy, Cerebral Palsy, and Blind were incorporated. 191 nondisabled adults completed the questionnaire.

Differences across social situations and among specific disabilities proved significant. Authors suggest future research should investigate a variety of disabilities and social situations, and the possible component underlying any situation-disability relationship.

Hahn, H. (1985), Toward a politics of disability: Definitions, disciplines, and policies. **Social Science Journal** 22(4), 87-105.

The author discusses the lack of attention by political scientists to the study of disability, and suggests positive contributions by this group. The three major definitions of disability - medical, economic, and socio-political - are discussed. It is noted that each discipline brings different values which contribute to the general definition of disability. The authors suggest that the socio-political perspective may best provide a foundation for alerting the expectations of the disabled and allow this population to seek adaptations to their needs. Hence the discipline's emphasis in understanding the consequences of a "disabling environment." The role of public policy in the shaping of any environment is stressed, and researchers are urged to encompass political aspects in the formulation of any further disability policy.

Hannah, M.E., & Midlarsky, E. (1985), Siblings of the handicapped: A literature review for school psychologists. **School Psychology Review**, 34(4), 510-520.

The authors review the existing literature regarding the effects of sibship with a handicapped child across a variety of conditions, and discuss potential intervention by the school psychologist for this at-risk group. The authors note that a child's handicap has the possibility for positive and negative impact on all family members, and researchers are now studying in earnest this implication for the siblings of disabled youths. This comprehensive review is divided into areas of concern fur the researcher interested in this complex relationship: family factors, characteristics of the non-handicapped sibling, characteristics of the handicapped child, and implications for school psychologists. The authors note that outcomes of research in this field vary widely concerning the degree to which adverse results may occur, and the nature of the adverse manifestations. Also, there is a paucity of research concerning what effects the handicapped child's actual sex, age, and type of disability have on the mental health of the sibling.

General conclusions drawn from the existing literature indicates that older females in small, low SES families who assume excessive caretaking responsibilities are at higher risk for psychological difficulties. The implication for the school psychologist is the need fur a more preventative, therapeutic role for siblings of the handicapped child, in addition to their more traditional role as diagnosticians.

Hannah, H.E., & Midlarsky, E. (1987). Differential impact of labels and behaviour descriptions on attitudes toward people with disabilities, **Rehabilitation Pschology**, 79(4), 227-238.

The primary goal of this research was to determine whether labels yield more negative reactions to persons with disabilities than do descriptors of the behaviour associated with the disability. The study investigated the way in which stimulus materials may influence attitudes towards disabled persons. The notion of "labeling" is discussed, and citations note authors who found respondents reacted more positively to unlabeled behavioral descriptions than to labels alone, or to descriptions of behaviour that are accompanied by labels. This study utilized the Attitudes Toward Disabled Persons Scale - Form O (ATDP, Yuker, Block, & Young, 1966) to determine this effect.

Findings suggest that the manner in which persons with disabilities are presented to peers affects responses to these persons. However, personality characteristics of peers were noted as influencing this outcome. Some conditions were found to yield a lower degree of acceptance when labels were used (i.e., neurosis). Conversely, some labels promoted acceptance (i.e., alcoholism, psychosis, prior conviction of crime). Authors note that only disabling characteristics were used in descriptors in this research and may have magnified negative reactions.

Jones, E., Farina, A., Hastdorf, A., Markus, H., Miller, D., & Scott, R. (1984). **Social Stigma: The psychology of marked relationships**. New York: W.H. Freeman and Company.

Social Stigma: The Psychology of Marked Relationships presents an incisive analysis of the stigmatizing process. This volume examines the social and psychological factors of relationships between those who are "normal" and those who are "deviant" in some fashion. Individuals with differences - the disabled, the disfigured, ex-mental patients, and ex-convicts- -are considered. One of the major strengths of this volume is the coordinated approach taken by this highly talented group of scholars. It represents the best single written compendium of the stigmatizing process completed to date in the United States.

Kaiser, S.B., Wingate, S.B., Freman, C.M., & Chandler, J.L. (1987). Acceptance of physical disability and attitudes toward personal appearance. **Rehabilitation Psychology**, 32(1), 51-58.

Personal appearance is an implicit but obvious component of interpersonal communications between persons with and without physical disabilities. Studies of interactions between these two groups would profitably consider self-perceptions of physically disabled persons as well as able-bodied individuals' perceptions of the disabled. The authors explored appearance management as a social-psychological variable in the lives of persons with physical disabilities. Data was obtained from a nationwide mail sample survey of 322 physically disabled university students. The overall hypothesis of a relationship between disability acceptance and attitudes toward personal appearance received little support. A tendency to regard appearance management as a form of compensation was reported as was a tendency to view one's physical disability as a central or salient aspect of personal identity. The data suggest that appearance management is regarded by some persons as a potential means for changing the social impact of physical disability, but is realistically expected to serve only a secondary role in impression formation. This study was done with university students, however it has very major implications for adolescents as they mature in social interpersonal relationships.

Kleck, R.E., & DeJong, W. (1983). Physical disability, physical attractiveness, and social outcomes in children's small groups. **Rehabilitation Psychology**, 28(2), 79-91.

The sociometric status of children with disabilities is examined. An integrated (70% black, 20% Puerto Rican, 10% white) summer camp was the setting for 60 boys (mean age 10.36 years) and 61 girls (mean age 10.13 years). Twenty-eight of these children (13 female, 15 male) were physically handicapped. Children with severe levels of functional and cognitive impairment were prohibited by the camp's admission policy.

Children were interviewed individually to determine acceptability as peers by the other children in their camp group (consisting of 5-6 children). Also, each child, having been photographed the first day at camp, was rated in terms of "attractiveness" by a panel of 12 boys and 12 girls from a neighbouring camp.

Results indicated that nondisabled children were better liked within their camp group than those obviously handicapped children. Also, highly correlated were physical attractiveness and social groups status, as those rated as being better liked were also perceived as being more attractive. Females made a larger contribution to this effect than their male counterparts. Also, able-bodied children with a higher social ranking were usually more accepting of disabled children as friends.

Phillies, M.J. (1985). "Try harder": The experience of disability and the dilemma of normalization. **Social Science Journal** 22(4), 45-57.

This author suggests that the process of normalization may no longer be the ultimate goal for persons with a physical disability. The recent ideological shift away from a traditional and stereotyped definition of success has helped redefine the role of rehabilitation for this population. The author interviewed 36 individuals with physical disabilities, ages 21-65. Discussed were the three ideological frameworks which influence these persons' ambivalence about being "normal:" the cultural notion of perseverance, the rehabilitation model, and the social minority model. Also, informants provided their dynamic approaches and definitions to their personal perspectives of success. The emphasis here was on an individual and context specific approach.

The author suggests that, as informants cite the implicit goal of the rehabilitation process is simply normalization, rehabilitation practitioners will need to respond to these changing ideologies by way of more individualized services in the future.

Pless, I.B., & Roghmann, K.J. (1971). Chronic illness and its consequences; observations based on three epidemiologic surveys, **The Journal of Pediatrics**, 79(3), 351-359.

Three epidemiological studies were utilized (National Survey, U.K., Isle of Wight, U.K., Rochester Child Health Survey, U.S.) to determine the psychological and social consequences of chronic physical disorders in children. These disorders are subdivided by type, duration, and severity of illness, and how they relate to secondary handicaps in the areas of educational achievement, behaviour, and psychological maladjustment. In all three of the studies, asthma was the leading single cause of illness, followed by sensory disorders (epilepsy) and central nervous system disorders (cerebral palsy). Comparable statistics were cited from each study to indicate that at least one.third and probably more of the chronic illness of childhood are likely to be permanent. The severity of the disorder, in terms of interference with ordinary daily activity, appears to have a direct influence on behavioral symptoms.

The authors indicate that most current health care systems are assigned for the child with acute episodic disorders and those children who were essentially healthy. "Management" as well as "treatment" is stressed for the comprehensive care required by chronically ill children. This entails thorough assessment of the child and family, as well as establishing baseline data in the areas of psychological development, schooling, and social relations of the child. The importance of utilizing lay professionals, as well as health care professionals, in this data gathering process in also discussed.

Richardson, S.A. (1976). Attitudes and behaviour toward the physically handicapped (Monograph). **The National Foundation Original Article Series**. 79(4), 15-34.

Steven Richardson is one of the premier authors writing in the area of attitudes toward childhood disability. Beginning with his early research in 1956, Dr. Richardson reviews over thirty years of research on attitudes and behaviour toward physically handicapped children in this article. Particular attention is placed on a review of personality and attitudinal research, value research with children, parental and child reactions to disability, and a comprehensive overview of future directions for such research in the area of physical impairment and psychosocial adjustment. Many of the ideas presented in the conference paper draw heavily from Dr. Richardson's writings and thinking.

Rosenbaum, P.L., Armstrong, R.W., & King, S.M. (1986). Improving attitudes toward the disabled: A randomized controlled trial of direct contact versus kids-on the block. **Journal of Developmental and Behavioral Pediatrics** 7 (5), 302-307.

Social interaction between able-bodied and disabled children is reported to improve children's attitudes toward handicapped peers. A randomized-factorial design evaluated the relative impact of two interventions, a buddy program, and the Kid's-on-the-Block (KOB) puppet program - singly and in combination. Outcomes included measures of attitudes, familiarity with disabled schoolmates, self-esteem, and parental attitude. The buddy program appeared to be successful as an intervention. The KOB program in combination with the buddy program produced a somewhat smaller, yet significant, impact as contrasted to the buddy program alone. Suggestions are offered by the authors for possible strategies to enhance interventions to teach social interaction skills to able-bodied children.

Seligman, M. (Ed.). (1983). **The family with a handicapped child: Understanding and treatment** New York: Grune & Stratton.

This text sets out to explore the family of a handicapped child from a number of perspectives, including external influences such as Legislation, the community, and the attitudes of health care providers. Also, various treatment interventions are discussed which address the psychosocial problems often faced by these families. The eleven chapters, each written by a practitioner in the field, cover four major areas of concern to this group: foundations; parents, the community, and service providers; family dynamics, and interventions. Stressed is the family's response to their exceptional situation, and the more recent notion that a family's reaction and adjustment to a handicapped child is not necessarily determined by the nature or severity of the child's handicap, but rather by the numerous factors which surround the family unit.

Servoss, Ann G.(1983). A physical minority: The disabled and mental health care. **The American Journal of Social Psychiatry III**, 2, 52-62.

The physically disabled are a group at-risk for differential treatment in all phases of life. Medical treatment, specifically psychiatric treatment, for this group is no exception. Historically, rehabilitation counsellors have been reluctant to seek psychiatric consultation for their patients. Conversely, some individuals who carry a "dual diagnosis" are often denied necessary treatment. This article highlights the unique problems of evaluation and treatment for the individual with a physical disability. Depression is cited as the psychiatric disorder which is witnessed most often in this population. However, persons with obvious physical disabilities may fall prey to various stereotypes which can lead to countertransference responses in the counseling situation. This may further contribute to altered body image and sense of self. The author stresses that patient, medical staff, and mental health care givers must work together to avoid this area of conflict and vulnerability for the person with an obvious physical disability.

Shukla, G.D., Sahu, S.C., & Tripathi, D.K. (1982) A psychiatric study of amputees, **British Journal of Psychiatry**, 141, 50-53.

Seventy-two amputees were monitored for psychiatric manifestations in the post-operative period. Sixty-eight patients were male and two-thirds of the cohort were in their first to third decade (81%). Fifty subjects had amputations resulting from accidents, and an almost equal percentage had upper and lower limb amputations. All patients were right handed, and almost two-thirds had right-sided amputations. Four-fifths of the study group were observed to have phantom limb phenomenon, with right arm amputees displaying phantom limb phenomenon and insomnia significantly more often. In addition, two-thirds of the study group had a psychiatric symptom of either depression, anxiety, crying spells, insomnia, loss of appetite, suicidal ideas, or psychotic behaviour. Two patients were diagnosed as schizophrenic, both right upper limb amputations.

Singleton, C.R. (1984). Indicators of social adjustment in juvenile amputees and implications for psychological intervention. **Dissertation Abstracts International**, 45(10), 3347-B.

The purpose of this study was to identify factors associated with the psychosocial adjustment of juveniles who have been congenital amputees since birth. The groups consisted of 19 amputees, ages 6-16 years, who had participated in the Juvenile Amputee Clinic of a regional children's hospital medical centre. Self-reports, parent and teacher ratings, and some medical personnel assessments were utilized to determine level of functioning in different social environments. It was noted that as the participation of adolescents in the clinic setting should be encouraged, mental health professionals should be included in the amputee clinic team to detect psychosocial conflict in the sometimes passive amputee.

Strohmer, D.C., Grand,S.A., & Purcell, M.J., (1984). Attitudes toward persons with a disability: An examination of demographic factors, social context and specific disability. **Rehabilitation Psychology**, 29(3), 131-145.

This research investigated two questions relevant to attitudes towards individuals with disabilities. The first question addressed the type of disability considered and the social context. The second addressed correlates of attitudes towards persons with a disability.

Study subjects were faculty and professional/nonprofessional staff of a large northeastern university. The Disability Social Relationships Scale (DSR) (Grand, et al., 1982) was utilized to measure attitudes towards persons with a disability. Subjects were also asked to complete three rating scales indicating the extent of contact they had with disabled persons.

Results indicated that attitudes do vary as a function of the interaction between disability type and social context, replicating the findings of Grand, et al. (1982). Results also support Yuker's contention that the rank ordering of preferences for disabilities may be situationally determined. The extent to which participants had previously had contact with disabled persons was the crucial factor associated with favourable attitudes.

Valliant, P.M., Bezzubyk, I., Daley, L., & Asu, M.E. (1985). Psychological impact of sport on disabled athletes. **Psychological Reports**, 56, 923-929.

The purpose of this study was to evaluate self-esteem and locus of control of disabled and non-disabled athletes. The Coopersmith Self-Esteem Inventory and Rotter's Internal-External Scale were used to obtain measures of each. The study group consisted of 139 physically disabled athletes, and 22 disabled non-athletes. Disabilities included amputations, blindness, cerebral palsy, and confinement to a wheelchair. Factors considered when predicting likelihood of group participation were place an individual was raised, reported level of happiness, living arrangements, type of institutionalization experienced, and level of education. The traditional role of athletic involvement as rehabilitation for the physically impaired person is discussed, as well as the role which self-esteem and locus of control play in group participation. Study results indicate that disabled athletes have a significantly higher self-esteem. exhibit greater life satisfaction and happiness are more externalised, and attain more education than their non-athletic counterparts. The authors suggest that psychosocial functioning of the physically disabled may be improved through participation in athletic events.

Vash, C.L., (1981). **The psychology of disability**. New York: Springer.

In the first volume of the Springer Series on Rehabilitation, Carolyn Vash discusses the psychology of disability. This work stresses the psychological principle that behaviour is a function of the individual interacting with the environment. Also, subjective experiences of the severely disabled figure prominently in this text, especially the psychological development in long-term living with a disability. Dr. Vash cites the two major issues of the work as devaluation and acceptance of the disability. Hence, the book is divided into two major sections: The Disability Experience- -the experiential world of the person with a disability; and Interventions - ways in which changes can be wrought by and for this population with disabilities. Dr. Vash discusses a comprehensive approach to a disability; the typical applied behavioral science embodied by professionals in the field, and the activist movement or the "politics" of disability. Dr. Vash states that she intended this work as a textbook fur students, as well as a general resource book for rehabilitation professionals.

Voeltz, L.M., (1980). Children's attitudes toward handicapped peers. **American Journal of Mental Deficiency**. 84(5), 455-464.

As children with a handicap are mainstreamed into neighborhood schools, it becomes increasingly important to understand the perceptions nondisabled students have of their peers. The purpose of the present study was to collect initial data regarding existing attitudes of children towards their severely handicapped counterparts. An attitude survey was administered to 2636 public school children in grades 2-7. Results indicate that children in this study were most likely to express a desire to interact with disabled peers if they were girls, in the fifth and sixth grades, and enrolled in a school with severely handicapped children. The author's cite studies which support the need for special activities designed to aid handicapped students in their newly integrated settings with nonhandicapped peers. The primary focus of this would actually be to enhance their status with nonhandicapped peers.

Wright, B.A., (1975). Social-Psychological leads to enhance rehabilitation effectiveness. **Rehabilitation Counseling Bulletin**, 214-223

Social perceptions which undermine situations of persons with disabilities are examined. Special problems of attribution, spread and the coping-succumbing frameworks, and of co-management and the helping relationships are addressed. Specifically, the author notes that the Rehabilitation Act of 1973 is a landmark which explicitly addresses the fundamental concepts of rehabilitation. Eleven recommendations are offered as suggestions for alleviating these potentially destructive situations. Suggestions include willingness to change the environment as well as the client's attitudes, and guarding against stereotyping people by their labels. It is stressed that counsellors should try to incorporate these procedures when working with a patient who is disabled.

Yuker, H.E., (Ed.). (1988). **Attitudes toward persons with disabilities**. New York; Springer Publishing Company.

Dr. Yuker brings together some of the best writers as well as researchers in the area of disability for this edited volume. Beatrice Wright clarifies major issues in understanding our negative bias toward those who are different and the politics of disability is clarified by the well-known rehabilitation psychologist, Professor Joseph Stubbins, of California State University. This chapter by Stubbins questions some of the fundamental ideas in our training of rehabilitation psychologists, our future research directives for those who are disabled, and the incongruity between governmental policies, client needs, and the myopia of professional researchers. All aspects of attitudinal research, including methodological evaluations, social psychological studies, and clinical implications are reviewed in this text.

13. Research priorities in cerebral palsy

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Our review on cerebral palsy (Rosenbloom, L. and Thomson, APJ. Pediatrician, 13.10.87), which provides an overall perspective on "the state of the art" in 1970, has been circulated. In addition to using the philosophy detailed in that paper I am grateful to other colleagues, working with me in Liverpool, who have shared with me their thoughts and studies on the epidemiology and treatment of cerebral palsy. These include Dr. Richard Cooke, Mrs. Theresa Cooke, Dr. Luis Lopez, Professor Peter Pharaoh, Dr. Teyrnon Powell and Dr. Michael Weindling.

Dr. Luis Lopez who is about to return to Costa Rica following four years of training in child development and child neurology in the United Kingdom has been a most creative source of ideas for my department. The studies and research projects on cerebral palsy which have come from our centre, on which I have collaborated are given in Fig. 1. They illustrate the catholicity of potential ideas that exist for cerebral palsy research at the present time:

Fig.1

PHARAOH ET AL, Trends in birth prevalence in cerebral palsy, Archives of Disease in Childhood, 1987 62 379-384.

PHARAOH ET AL, Effects of birth weight, gestational age and maternal obstetric history on birth prevalence of cerebral palsy? Archives of Disease in Childhood 1987 62 1035-1040

POWELL ET AL, Cerebral palsy in low birth-weight infants II Spastic diplegia? Associations with fetal immaturity? Developmental Medicine and Child Neurology 1988 30 19-25.

WEINDLING ET AL, A random controlled study to determine whether early physiotherapy reduces disabilities in children who are likely to develop cerebral palsy. On going research study.

Fig. 2 indicates the major headings under which I shall give my personal view of cerebral palsy research priorities.

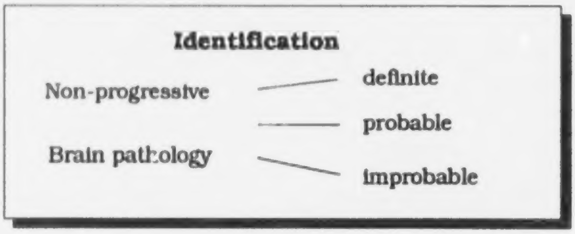
Fig. 2

Identification
Primary Prevention
Early Detection
Comprehensive Assessment
Remediation

Identification

At first sight it might seem that the first "identification" is both out of place and out of context. Yet knowing which children have cerebral palsy and perhaps more importantly which do not, is fundamental to any rational clinical scheme or service of prevention and management. Moreover, at the micro level of understanding individual children and helping their families, a knowledge of pathology and pathogenesis is of great importance.

Fig. 3



The first question to be asked is - Is it definite, probable or improbable that we are dealing with non-progressive brain pathology? There is more to this broad question than that of defining which children have cerebral palsy. In these litigious times, so long as we accept that the non-medical professions have every right to focus our minds, the problems of whether perinatal hazards are the cause of neurological deficits in individual children, needs to be scrupulously addressed. I shall return to this point at the end of this paper.

At a scientific level however we then have to go further and we have to use not only clinical but also investigative data to determine to which of the categories individual children should be assigned. The recent delineation of Rett's syndrome as a progressive disorder, with a hitherto unidentified biochemical deficit, indicates how a small but possibly increasing number of children assumed historically to have cerebral palsy, may well turn out not to be so affected when relevant investigations become available. It follows that in modern practice suitable schemata of investigation are an essential component of both research and clinical management. Time does not allow a review and critical appraisal of the increasing number of investigative methods - radiological, neurophysiological and biochemical that are available. But we need protocols that are prospective and multi-centred, that identify potential at risk and control populations, that use rational investigative schemes, that if need be can be flexible and where appropriate can be linked to management studies and programmes. One has to be critical of both United Kingdom and North American neonatal and paediatric neurological practice in this context. Our oncology colleagues seem to have received this message long since.

If this approach were adopted a number of advantages would accrue. Firstly, our epidemiological data on incidence and prevalence would be more accurate, secondly we could link investigative and clinical findings with some precision and thirdly, within the context of cerebral palsy, the reasons for the particular vulnerability of the pyramidal tracts, the extrapyramidal system and the cerebellum and its connections could be explored with a view to limiting the ultimate amount of damage.

It is interesting to speculate also that, so far as the perinatal causes of cerebral palsy are concerned, that the pathological changes progressively occurring in association with hypoxia and vascular abnormality and that lead to cystic degeneration are in fact progressive albeit over a short period of time, rather than non-progressive. Could it be that modern studies of the cause of cerebral palsy will lead, after 25 years or more, to a re-definition of this non-progressive syndrome.

Fig. 4

Identification

Need for prospective,multi-centred controlled clinical + investigative studies

Primary prevention

Returning now to the second of my major headings, primary prevention, it will be understood why I considered it important to identify earlier what it is precisely we are discussing. In pathological terms we are saying how can we prevent the pyramidal tracts and other neuromotor systems suffering damage in the first place.

A distinction has to be made at this stage between specific and non-specific measures that can be applied pre-conceptually and later during early pregnancy. Here it has to be noted that the availability of specific measures is a generation or more away; perhaps the mapping of chromosomes has something to offer here for the few genetically determined causes of cerebral palsy.

By contrast it is to me a matter of considerable regret that we cannot precisely identify either which non-specific factors seem to so adversely affect the uterine environment as to cause cerebral palsy, or know how the relevant parts of the brain are damaged. Is it a matter of chance, of the organism or the timing in pregnancy for example, that maternal rubella or cytomegalovirus affects the pyramidal tracts. Again we talk glibly of better care of women during early and mid-pregnancy in terms of nutrition, the avoiding of tobacco and alcohol and the maintenance of good health generally and yet there is really no evidence that this approach diminishes the number of children with cerebral palsy. And yet we do not know whether this approach is not generally undertaken because it is wrong, or socially unacceptable or whether it is our measuring instruments that are unsuitable by perhaps being too blunt. In this area of primary prevention, we must move a step away from epidemiological studies and provide more clinical models.

Much the same has to be said about perinatal factors that are thought to cause cerebral palsy. How do immaturity, asphyxia, trauma, infection, nutrition and poor temperature control alone or in combination damage babies brains. Until research methods provide us with models that can take into account the plasticity of the human nervous system and answer these questions, we must remain unsure as to how much of the investment that developed countries can be putting into perinatal care is wholly relevant. Although not strictly comparable, the impressive recovery that so many head injury children make after road traffic accidents, reinforces the concept that even when stressed, the brain has considerable powers of recovery. Does this apply, and if so how during the perinatal period?

In prevention therefore the research priority is to provide suitable clinical models that can give us some of the answers to aetiology.

Early detection

To change tack, I now want to consider some aspects of the early detection of children with cerebral palsy. This first research priority in this area is to justify early detection measures themselves. It is perhaps an article of faith that early detection is "a good thing" and that in some way it does help both the children and their families or both. Hard evidence that this is so is however difficult to come by. Our own study into the effects of early physiotherapy on high risk infants may go some way towards answering this question. In this we are looking at newborn infants who are identified as being at high risk of developing cerebral palsy as a result of their cranial ultrasound and somatosensory evoked cortical responses. Half of such children are then randomly allocated to early physiotherapy and the remainder follow our present practice and are offered physio- therapy only if and when motor development is delayed or abnormal.

As well as carefully monitoring the development of the children in both groups we are looking also at the reactions and responses of the parents.

Given that this study will hopefully identify children when presymptomatic and follow them up throughout childhood, it could be that the family adjustment measures used by our psychological colleagues would be of particular interest and significance in justifying early detection. So also of course will the information on the benefits or otherwise of early physiotherapy but this is a point that I shall be returning to presently.

If however we can accept for the sake of argument that early detection is beneficial then relevant measures that can be applied to appropriate populations need to be instituted. I cannot help feeling that studies such as that from Oxford which looked at non-walking eighteen month old infants are an example of closing the stable door rather too late, and once more, this is a task we should be attempting to undertake prospectively. There are a number of possible ways that this could be done.

Fig. 5

Early Detection
Developmental screening programmes
At-risk follow up
Identification by parents

Firstly, a population developmental screening programme aimed **specifically** at looking at motor delay and deviancy has much to commend it. Generally speaking developmental screening programmes have in my view attempted to do too much for too many with imprecise methods, imprecise aims and wooly compromises that have rendered them fair game for destructive criticism and disregard.

It could reasonably be argued that it would be logical to identify:

a) the type or pattern of abnormality to be detected; this implies a knowledge of the normal range of skills to be expected for children of particular ages,

b) the measures to be used in their detection,

c) the personnel and other resources needed for such an identification programme

d) as discussed above management measures that are available to alleviate the disabilities once detected.

With the exception of the availability of screening for hearing loss by distraction testing and I shall not digress to discuss the major difficulties in implementing that programme, motor abilities and disabilities are the only developmental parameters that fulfill the criteria for a developmental screening programme as defined above. Certainly mental handicap, visual problems and emotional disturbances do not.

I would suggest therefore that using primary care professionals, for example community nurses, that the population of children who at appropriate times corrected for their gestational age in the first year have tone abnormalities, persistence of primary responses or delay in head control, reaching, sitting, crawling or shuffling, standing and walking could and should be identified. What has never been done before is to gear such a programme specifically to motor disorders in terms of both the timing and orientation of the examinations. Any spin off in finding other developmental disorders would be valuable but not the primary aim of the study which clearly would have the advantages of being both feasible and focussed.

Two other studies concerned with early detection that contrast with this methodology could also be of interest. The first involves more systematic follow up of children discharged from neonatal special care units. Knowing these are a vulnerable population there may well be some mileage in allocating physiotherapy resources for their clinical motor assessment. One is not desperately impressed with current services for clinical follow up of these infants and given that significant motor abnormalities might initially be subtle their detection might well be facilitated by appropriately focussing the attention of examiners.

The alternative approach, and one that I have long advocated as a principle, is that of equipping parents to be the primary screeners of their children for developmental difficulties. Here motor abnormalities would have to be looked for within the context of the children's general development and behaviour. The evidence hitherto for what it is worth is that parents are not particularly good at picking out children who have motor problems as opposed to language learning and behavioral difficulties. If this turns out to be the case however then one wonders if it could be changed by a parent education programme commenced antenatally or even pre-conceptually.

Certainly if we could equip parents, within the context of promoting their children's health overall, to be able to be in the front line of detecting their disabilities, this could only be advantageous. A first step would be the extension of parent held child health records to make them more comprehensive and more central to children's care, perhaps in some way by linking them to social security or similar benefits. I would certainly commend this approach as a research option.

Comprehensive assessment

Evaluation of the likely multiple disabilities of children with cerebral palsy has medical, educational and social parameters. I find it disappointing, not to say remarkable therefore that systems for comprehensive assessment have never really been established or published. Let me illustrate some of the areas of difficulty. First, is a better yield of information obtained from centralising all the assessments at child development centres or other handicapped children centres, or would we do better by supplementing more peripheral community services and resources by putting in to them any additional expertise that is needed.

Fig. 6

Comprehensive assessment

centralised / outreach
brief / prolonged
combined / sequential

Second, is there a better yield from the sort of assessments that go on for three weeks or more, often in a nursery situation or can appropriate amounts of information be obtained in much shorter periods and thirdly, and associated with that, is a model in which the many concerned professionals see the child and family sequentially more productive than on involving combined assessments.

Moreover, and so far as the motor deficit is specifically concerned, there really has been minimal research concerned with recording, collating and evaluating data. It is not surprising therefore that it is so difficult to judge the effectiveness of physiotherapy regimes. As a result it would be helpful to see an extension of the motor deficit descriptive scheme advocated by Evans and her colleagues so that it or something similar could be developed for use by physiotherapists as an assessment tool.

Remediation measures

Fig. 7

Remediation research needs

- study designs
- technological resources
- media involvement

In discussing remediation for children with cerebral palsy I propose to illustrate the difficulties and research opportunities by concentrating on two subjects only. One, superficially at least specific? and the other more general.

The first is a comment on the current fashion which I share, for ankle-foot orthoses (AFO's). In our centre my orthopaedic colleagues, physiotherapists and I are using these extensively to help correct the equinus foot postures of children with cerebral palsy. We think they work. By that I mean that properly and skillfully measured manufactured and fitted AFO's produced by our own orthotic service and applied to both feet of diplegic or hemiplegic children when there is a bilateral or unilateral dynamic equinus interfering with locomotion seem in most affected children to make walking easier and more cosmetic. We have also attempted to prove this using gait analysis techniques, but in our centre these are not sophisticated enough. I do not therefore see any alternative to investment in appropriately equipped and clinically directed gait laboratories for evaluating appliances such as AFO's or orthopaedic surgical procedures. These need to be within the context of the availability of paediatric, orthopaedic, physiotherapy, orthotic and other technical services that work well together. In the absence of this interdisciplinary working there is considerable danger of the technical tail wagging the clinical dog.

Finally, I come to the general point of children with cerebral palsy. It is inevitable that sufferers from incurable conditions become prey to enthusiastic or even bogus treatment claims. The current United Kingdom enthusiasm for conductive education is a case in point. In the light both of past history and the present day, it is clear that discussion and research on how to promote neuro motor progress and to correct neuro motor abnormalities is essential, but the research needs to be funded, and research protocols and their prosecution need to be vigorously regarded. Thus following on from my earlier point about the need for description and analysis of the motor deficit in individual children, it follows that treatment schedules need to be equally carefully analyzed, that associated factors whether these be medical, social or emotional need to be taken into account, and most importantly, treatment goals should be identified in detail. I would suggest that computer models for such a complex approach are feasible and it would in my view be helpful to attempt to apply them.

This leads me on to my last point. It seems to be currently fashionable for the lay public and the media to be skeptical of what doctors can offer and specifically in this context there is the implication that the medical and related professions are negativistic in their approach to handicapped children making progress. I think the onus is on us therefore to work with communication professionals and the media generally to rebut this. This implies honest evaluation of our own work, appreciation of the limitations of our knowledge and resources, an awareness of communication technology and an appreciation also of the needs and weaknesses of the media. What I am asking for is research on how the medical and related professions on the one hand, and those concerned with the media on the other can profitably work together to illustrate and highlight problems including those of resources, treatment needs and possible solutions.

The research ideas for cerebral palsy that I have suggested in this paper range from the very pure to the wholly applied. They have included comments on diagnostic measures, epidemiology, identification, evaluation and treatment. None are mutually exclusive or contradictory. Some need a reorientation of approach, others cooperation with colleagues and many need resources. Above all we need a perspective on where we are and where we are going. It is us, rather than others who should contribute to decisions on both of those.

14. Spinal dysraphic states

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"I had a figure of the divided spine, however malformed, neatly engraved in copper; let the surgeons not unprudently open such a swelling but force them to see the shame this would undoubtedly bring upon them."

Nicholas Tulp, 1642

The art of medicine in the 17th century as described by Nicholas Tulp has been expanded and strengthened by a degree of scientific medical advances undreamed of even 50 years ago. However, there remain several dilemmas associated with the understanding and treatment of the dysraphic spine of which spina bifida remains the most common condition requiring medical treatment.

First it must be recognized that the medical and surgical treatments designed for congenital spinal defects will always by definition remain palliative and not curative. Secondly, to optimally treat a disease process in the most appropriate fashion, one must have a firm grasp of the natural history of the disorder. In the field of spina bifida therapy, new treatment modalities continue to be introduced which, while initially offering clinical promise, may at the same time introduce new variables into the equation of the history of the disorder. A case in point is the ongoing orthopaedic controversy regarding the optimum surgical treatment of the paralytic hip in spina bifida for which no truly long-term outcome studies are available. Thirdly, there remains much confusion regarding the exact pathophysiology of the neurologic lesion itself. Early recognition of the upper motor neuron component in as many as a third or more of cases was frequently overlooked or ignored in past decades, only now to be again recognized to be a significant factor in the variable clinical course some children exhibit. Lastly, there is the major economic dilemma facing all health-care providers today, namely, allocation of health-care resources. Since the introduction of an effective ventricular shunt by Holter in 1957, there has been an ever increasing group of survivors, many of whom are now entering their adult years. We have only preliminary knowledge of the medical and social needs these patients will require of society in an era of increasing longevity and finite medical resources. At the same time, it can be effectively argued, however, that the present adult survivors may not accurately reflect the projected outcome of younger patients now receiving "improved" therapy or treatment based on newer medical knowledge.

The ultimate answer to the "cure" or "control" of spina bifida remains locked in the challenge to reduce the incidence of the embryologic defect. Divergent hypotheses of herniation of a previously formed neural tube, first postulated by Morgani, Virchou, and Cleeland versus the failure of closure proposed by vonRecklinghausen serve as models for continued teratologic research. The report of Osaka and colleagues(11) in 1978 is an example of the wealth of information that can be gathered from a systematic analysis of aborted fetal material. A closer collaboration between teratologists, neuropathologists, and those clinicians involved in the care of females in high risk pregnancy centers would be invaluable. The Congenital Anomalies Research Unit in Thornbury Hall, Sheffield, England, entirely supported by private donated funds, is a prime example of such a unit where much data was discovered in the 1960's and early '70's.

The departments of Genetics at the University of Connecticut and Yale University have collaborated on a joint statewide program to provide genetic counseling and obstetrical and perinatology care for all women at risk within the state of Connecticut. Additional support to systematically analyze these specimens combined with the advanced genetic counseling data may prove rewarding given the rarity of this congenital anomaly.

Much interest has been generated by Smithells(16) and others concerning the issue of nutritional deficit and vitamin supplement therapy, especially folic acid. One should pause, however, to reconsider that one of the factors leading to investigating the concept of nutritional factors was the long-recognized relationship between the increased incidence of neural tube defects in the lower socio-economic classes. Nevin(10) reported an interesting observation at the 31st Annual Meeting of the Society for Research into Hydrocephalus and Spina Bifida in 1987. With the introduction of supplementary vitamin therapy in Belfast, Northern Ireland, the abnormally high endemic rate of neural tube defects was reduced but not eliminated. Rather, the post-treatment incidence approximated the more commonly reported incidence of 1:1,000 births. Much attention has been directed to various Celtic populations in the United Kingdom and Ireland. However, few investigations have addressed the newly emerging Hispanic populations in North America. We have been impressed by a relatively large number of affected babies of Hispanic origin requiring treatment in the state of Connecticut for which little or no genetic or demographic data is available.

Guthkelch(4) reviewed in great detail the complexities surrounding adequate treatment of a condition for which the underlying neuropathology is either poorly understood or misunderstood. Confusion still exists in the clinical setting when one fails to comprehend the basic differences in embryologic development between the growth of the neural tube cephalad to the posterior neuropore and the development of the tail bud by a process of canalization of the caudal cell mass. The development and ultimate clinical prognosis for a child with a lumbar lipomeningocele will frequently be different from that of a child with a similar level myelomeningocele with hydrocephalus. On the other hand, the child with a pure lower motor neuron paraplegia, regardless of the level of involvement, will often be substantially different over a long period of time from a child with an identical level showing an upper motor neuron lesion. As clinicians, we are only now beginning to realize the major late-term clinical implication of these factors.

How can the neurologic lesion be most accurately diagnosed and defined? Newer imaging techniques, including metrasimide enhanced CAT scans, magnetic resonance imaging(MRI), and ultrasound, offer great potential. However, the definitive value of the numerous short-term reports that identify lesions and often compare the accuracy of other imaging modalities must, in my opinion, be coordinated over time with the clinical course of the patient with further growth. These techniques not only reveal newer insights into previously obscure pathologic conditions but also they force us to reconsider established definitions of normal versus pathologic states. Recent MRI studies have shown, for example, that the tonsillar ectopia, previously associated with Arnold Chiari Type 1 malformations, has a normal range of 1 millimetre above to 5 millimeters below the foramen magnum in otherwise normal subjects(2). Prospective imaging studies should evaluate not only the thoracolumbosacral regions but the craniocervical regions of the spine as well.

Electrodiagnostic studies have shown considerable recent promise. Previously, Faradic stimulation at the motor points in paralyzed limbs suggested by the Sheffield group, provided no information about the peripheral nerve, root, or cord proximal to the point of stimulation. Cystometrographic analysis of the bladder function is an area of study that holds great promise now and in the future. However, for its greatest value, multidisciplinary investigation should be encouraged to combine the urodynamic findings of the urologist with the clinical and imaging data of the orthopedist and neurosurgeon to better define the precise nature of the neurologic deficit.

The surgical aphorism that to improve outcome the surgeon must either devise new operations or be more selective in the performance of existing procedures is most appropriate in the field of myelodysplasia. Nowhere is our need greater to accurately assess the neurologic lesion than in the review of orthopaedic literature concerning the treatment of paralytic hip disorders. Ralis(13) summarized the current diversity of opinion in the symposium on hip disorders at the 30th meeting of the Society for Research into Hydrocephalus and Spina Bifida. Improved results will only be forthcoming with precise, accurate assessments of the neurologic level. Previously successful orthopaedic procedures for the treatment of poliomyelitis, a lower motor neuron disease, have been less effective in the treatment of spina bifida. In a large part, this is due to the absence of protective and proprioceptive sensation as well as the previously recognized high association of upper motor neuron lesions. Menelaus(9) provides a cogent plea for the well-planned, realistic treatment of these children based upon realistic rehabilitation goals.

The technology of spinal instrumentation and surgical techniques has reached a greater degree of success than was thought possible 15 years ago(1). Problems remain, however, in assessing the child with reflex motor activity, asymmetric motor levels, and the small child with retarded growth and poor weight. More collaborative study with neurologists, pediatricians, and nutritionists is needed to promote optimum growth and reduce perioperative complications in these children.

The field of orthotic management in myelodysplasia is dynamic innovative, and expensive. In the course of searching for the ideal orthosis, several factors should be recognized. The child in the first decade of life is the most appropriate subject for the upright experience, for not only mobility per se but for the intangible benefits of growth and development. These "intangibles" bear closer inspection by occupational therapists and physical therapists and developmental experts. Little objective data has been obtained to substantiate the costs involved in the delivery of complex systems which require frequent, expensive replacement due to growth. The law of mass in the growing child is often overlooked. Briefly stated, a person's mass or weight increases as the cube; however, the strength increases as the square, leading to the inexorable conclusion that with growth, weight relatively exceeds strengths. Further investigation of the metabolic demands on the paralytic child using mobility devices will be valuable in establishing better guidelines to the delivery of upright walking aids.

Mobility remains the major objective of the child or adult with paraplegia. Electrical stimulation, currently popular with adults and adolescents with acquired traumatic paraplegia, has achieved modest success in persons with previously normally developed neuromuscular systems who were subsequently rendered paraplegic. To apply similar, expensive techniques to children with congenital disorders who did not previously walk with normal neuromuscular control is not likely to have a high benefit/expense ratio. Wheelchair design improvements in materials, weight, and adaptations for growth appear to be more realistic goals at this time. Future applications of electrical stimulation in select patients with congenital paraplegia maybe applicable with further miniaturization and refinement in the technology(12). Effective utilization of this technique however, will require very close cooperation between the rehabilitation engineer, the therapist, and the clinician who promote improved visual/motor coordination skills as well as the prevention of progressive deformity due to motor imbalance and growth. The measure of a society's achievement has by many philosophers been best judged by how a society provides for its less fortunate or least physically endowed members. The last 17 years since Lorber's report of prognostic criteria for possible selection for treatment(9) has seen many re-evaluations of not only the ethics of treatment as noted by the Ethical Working Group in the 1985 Lancet report(15) but also a critical appraisal of what care to render to newborns(3, 8). If one attempts to reconcile these already difficult decision-making problems with the suggestions by Guthkelch(4) that a well-controlled trial of immediate versus delayed closure be undertaken, one cannot but examine the need for a more centralized referral policy for the initial management of such children. Given the relative rarity of the condition, there would be the probable benefit of a greater number of patients available for more accurate analysis. A potential detriment might be the risk of subjecting patients to arbitrary biases of the particular treating unit. Multicentre, collaborative efforts already established for the treatment of childhood oncology might serve as models for such a study.

The variants of spinal dysraphism, such as the tethered cord syndrome, provide a very favourable area for study, even though the incidence of such anomalies is even less common. The recently published report on the first Stonwin Conference(5) provides an exceptional case in point in which leading specialists in the field of neurosurgery, radiology, and urology collaborated in the collection of valuable basic science and clinical material. Such joint efforts should be encouraged in the future to further explore the relevance of "tethering" in cases of spina bifida cystica as well. It must be emphasized that longitudinal study of affected individuals is crucial to the evaluation of any treatment modality. Recently, unpublished data from the Newington Children's Hospital on long-term follow-up analysis of selected patients with lipomyelomeningocele revealed an unexpectedly high incidence of serious late foot deformity, in some instances leading to amputation. Based upon current knowledge of the often subtle manifestations of altered neurological function secondary to compression or traction, many adults with lipomyelomeningocele demonstrated disabling deformities of the insensate lower extremities. Finally, we are all becoming aware of the emerging population of young adults with variable degrees of disability. In a large part, the life-threatening crises of overwhelming sepsis, shunt malfunction, and crippling orthopaedic deformity can be ameliorated or reversed with modern medical or surgical techniques. The problems of chronic renal insufficiency, mental retardation, obesity, lack of motivation and self-esteem, and the inability to become competitive and productive members of adult society are becoming self-evident. The greatest threat we face as specialists in the study of a multi-faceted disease, is to lose perspective of the whole patient. There are valid reasons for re-examining the current state of the young adults with myelodysplasia. There are very few long-term analyses of adults with spinal dysraphism. Clinical psychologists and social workers can be of invaluable assistance in clarifying the current problems of the disabled adult with myelodysplasia and assist us in placing our treatment objectives into better focus for the whole patient, not only for the immediate presenting problem but also for the optimum attainment of a productive adult existence. The field of sports and recreational programs for the handicapped(14) offers one area of positive support for the disabled. The opportunities provided through such programs deserve wider exposure in the treatment clinic, school, and community.

The spectrum of spinal dysraphic disorders is broad and the degree of impairment highly variable. Most puzzling of all is that the underlying cause of the disorder is elusive. I have attempted to pose what I believe to be some of the most pressing questions that confront physicians attempting to treat the patient with spina bifida. Currently, we are at a crossroads in the knowledge of spinal dysraphism, realizing some of the long-term implications of the disorder today, yet lacking a clear understanding of its cause. This critical junction is perhaps best expressed in the words of Victor Hugo(6) : "Where the telescope ends and the microscope begins, which of the two has the grander view?"

References

1. Banta J.V., The evolution of surgical treatment of spinal deformity in myelomeningocele. Zeitschrift fur Kinderchirurgie(suppl) , 1:10-12, 1987.

2. Barkovich, A.J., Sherman, J.L., Citrin, C.M., Wippold, F.T., M.R. of postoperative syringomyelia. Am J Nucl Radiol, 8(2): 319-327, 1987.

3. Gross. R., Newborns with myelodysplasia - The rest of the story. Editorial. N Engl J Med, 312(25): 632-1634, June 20, 1985.

4. Guthkelch, A.N., Aspects of the surgical management of spinal myelomeningocele; a review. Dev Med Child Neurol, 28:525-532, 1986.

5. Holtzman, R.N.N., Stein, B.W.,(eds) The Tethered Spinal Cord (Report of the First Stonwin Conference), New York, Thieme-Stratton, Inc, 1985.

6. Hugo, Victor, Les Miserables, "Saint Denis," Bk III, Ch 3. Tr by Wilbour C.E., Modern Library.

7. Lorber, J., Results of treatment of myelomeningocele. An analysis of 524 unselected cases, with special reference to possible selection for treatment. Dev Med Child Neurol, 13:279-303, 1971.

8. McLaughlin J.F., Shurtleff D.B., Lamers J.Y., et al: Influence of prognosis on decisions regarding the care of newborns with myelodysplasia. N. Engl J Med, 312(25): 1589-1594, 1985.

9. Menelaus, M.B., Orthopaedic management of children with myelomeningocele; a plea for realistic goals. Dev Med Child Neurol(Suppl) 37:3-11, 1976.76.

10. Nevin, N.C., personal communication, July 1987, Belfast, N. Ireland.

11. Osaka, K., Tanimura, T., Hiroysima, A., Matsumoto, S., Myelomeningocele before birth. J. Neurosurg, 49:711-724, Nov 1978.

12. Peckham, P.H., Keith, M.W., Freehafer, A.A., Restoration of functional control by electrical stimulation in the upper extremity of the quadriplegic patient. Current Concepts Review, J Bone Joint Surg, 70A:144-148, 1988.

13. Ralis Z.A., The outcome of treatment of hip problems in spina bifida. Zeitschrift fur Kinderchirurgie.(suppl), 1:56-60, 1986.

14. Sports and Recreational programs for the Child and Young Adult with Physical Disability. Proceedings of the Winter Park Seminar, April 11-13, 1983, Document #993-883, The American Academy of Orthopaedic Surgeons.

15. The prognosis for babies with myelomeningocele and high lumbar paraplegia at birth. Commentary by an Ethical Working Group, Lancet, Nov. 2, 1985, pp 996-997.

16. Smithells, R.W., Sheppard, A., Schorah, C.J., Seller, M.J., Nevin, N.C., Harris, R., Read, A.P., Fielding, D.W.: Apparent prevention of neural tube defects by periconceptual vitamin supplementation. Arch Dis Child, 56: 911-918, 1981.

17. Tulp, Nicholas Observationum medicarum libri tres-cum acreis figuris apud ludovicum. Elzvirium, Amsterdam, 1642.

Spinal dysraphic states

Discussion

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Dr. Banta has given us an excellent assessment of the major clinical issues and areas of research emphasis with regard to neural tube defects. I have touched briefly on some of the same ground in discussion of previous sessions at this conference. In this discussion, I plan to add some of our favorite Seattle harmonies to Dr. Banta's tune, suggest some new melodies and play a reprise on the theme of qualitative issues in research support.

Our successes in advancing the management of neural tube defects in Seattle have come as a result of a nearly 30 year effort by a single research and clinical care team (Shurtleff et al, 1986). I can list the accomplishments without being immodest because I have been on the scene only ten years and am more a student than an originator of our work. Some of the conditions that have allowed such an effort to flourish are instructive because they are reproducible. It is also possible to learn from our mistakes. Anyone looking to invest funds in research on a chronic disorder or disability should look first at the energy and commitment of the primary investigator since any long term project is going to be faced with obstacles, boredom, and competing priorities. Any of you who know Dr. David Shurtleff understand that he is the mainspring that drives our effort. Long term research is not for the faint hearted. Our focus encompassed the disorder, the impairments, the disabilities and the handicaps that make up "myelodysplasia" from the beginning. A multipronged research and clinical care thrust has made the program robust enough to withstand the buffets of academic and funding fluctuations and attract people with diverse talents to various aspects of the enterprise. This is a reproducible characteristic. We have been fortunate to have core support from the March of Dimes for many years. The steady support has allowed us to maintain a logical sequence of investigation even though the dollar amount of the support has not kept pace with expansion of the program. I hope that our success will reassure some funding sources that long term investment in a single group of researchers can pay off. The caution to add is for researcher and funding source alike to avoid complacency and cronyism at all costs. I believe that another important factor in our success has been that we have maintained vigorous involvement in teaching and have remained heavily involved in direct patient care. The former has given us a steady influx of fresh, challenging talent in many disciplines. The latter has garnered us the long term trust and relevance necessary to keep an interested patient population willing to let us learn the successes and failures of intervention. New hypotheses arise constantly from the rich clinical milieu to drive our research effort.

Dr. Banta has been modest about the contributions of his group. They have published a number of important papers in the orthopedic literature (Banta. 1987), and they have been leaders in the use of modern gait analysis (Gage. 1984). Their innovative gait lab is the first to be entirely automated and propels the study of the locomotion of physically disabled children from a cumbersome research instrument to a tool that has essentially "on-line" research and clinical application. The manner in which the lab was developed is instructive for this conference because it represents another model of collaboration. As I understand it, a private corporation entered into a joint effort in order to tap into the knowledge of biomechanics possessed by the clinical staff. The private corporation provided engineering and computer expertise, and had some research goals in industrial robotics that dove-tailed with the clinical research interests at Newington. Joint research is a model that has not been used much in medicine but has had wide spread application in the other sciences. Joint ventures are proliferating in the recombinant DNA technology field and venture capital is there for the right project. As with any model of research funding there are hazards in this approach, commercialization and exclusivity are the major concerns but are nothing new when measured along side the pharmaceutical industry.

Computer technology of a modest sort has been the key to the development of the extensive clinical data base that we have in Seattle. It has gone through several revisions as the science of information management has evolved. At present our system is built around commercially available software running on IBM-AT microcomputers. Dr. Shurtleff's programmers have devised a very valuable data management application of R:BASE System V that has been dubbed the Pediatric Data Management System (PDMS). PDMS has been shared through the International Myelodysplasia Study Group IMSG with more centers than I can remember and is in a state of continuous development. Using this system, we and our IMSG partners collect standardized clinical data some of which is common to several centers and some of which is unique to a particular centre. Data is collected for two major uses: a clinical register from which one can select subgroups for clinical review or initiation of new studies and sets of data to answer prospectively framed research questions. We have found that it is necessary to dedicate staff to both the collection and entry of data. While much of the data is gathered while children are being seen for clinical purposes, we have not been able to rely on the clinician to gather systematic data and still care for the child. We have either hired specific staff or depended on the efforts of graduate students who derive their thesis material from our study population. A great deal of effort has gone into validity and reliability checks and the use of closely written data collection protocols. We have also learned that the only data that stays reliable, even with this effort, is that which is important to someone because it serves a present research or clinical need. These points may seem a bit mundane but they are important for anyone considering a clinical research effort that involves long term data collection. Multicenter data collection is even more complicated but is essential to dealing with some of the uncommon problems of myelodysplasia.

Now, I would like to reiterate and perhaps add to Dr. Banta's list of areas in need of further research in myelodysplasia. In outline form they are as follows:

Disorder

1. Basic pathoembryology (Lemire et al, 1975)
2. Differentiation of primary and secondary neural tube defects (James and Lassman, 1981).

Impairment

1. Pathophysiology and treatment of central ventilatory dysfunction (Hesz and Wolraich, 1985)
2. Contribution of brain dysfunction to mobility problems in addition to spinal paralysis
3. Management of tethered cord and hydro myelia
4. Management of kyphosis (Banta, 1987).

Disability

1. Development of adaptive equipment
2. Role of ambulation and adaptive equipment in independent mobility
3. Adult function
4. Development of self-esteem and independence.

Handicap

1. Efficient organization of services
2. Societal policy research
3. Anthropologic aspects (e.g. wheelchair access in the urban and rural environments)

Finally, Dr. Banta's presentation stimulated me to think not only about the quantitative side of research, but to consider some qualitative issues. Again, I will touch on them in telegraphic form expecting that the more viable issues will become clear in group discussion. I propose that further research in myelodysplasia has need for the following:

1. Continued innovation and risk taking in what is still a young field.
2. Big and small ticket technical development like the Newington gait lab and better bowel management curicula
3. More longitudinal research support for more centers to allow maintenance of stable research teams with a consistent focus. It is destructive to be required to change directions with the whim of research fads.
4. Support of innovative clinical care which yields loyal, interested patient population in which one can demonstrate applications from the lab and from which one derives new hypotheses in real time.

Most of these needs run somewhat counter to the structure of major funding programs and have been the particular domain of private foundations. Given the tasks that still lie ahead, I believe that there needs to be better coordination among all sources of funding, public and private, profit and nonprofit, to efficiently support pilot projects by new investigators, capital investment in the tools of research. Best-practice clinical care in research environments, and programs that are sufficiently endowed with long term support to carry out 20 to 30 year research agendas. Such programs need to be structured to keep research teams at the cutting edge.

References

Shurtleff, D.B., et al. Myelodysplasias and Exstrophies; Significance, Prevention. & Treatment. Grune and Stratton. NY. 1986.

Banta, J.V., The evolution of surgical treatment of spinal deformity in myelomeningocele. Zeitschrift fur Kinderchirurgie (supp) 4: 715-725, 1984.

Gage, J.R., et al. Pre- and post-operative gait analysis in patients with spastic diplegia. Journal of Pediatric Orthopedics 4: 715-725, 1984.

Lemire, R.J., et al. Normal and Abnormal Development of the Human Nervous System. Harper and Row, Hagerstown, Md. 1975.

James, C.C.M., and Lassman, L.P., Spina Bifida Occulta. Academic Press. London, 1981.

Hesz, N., and Wolraich, M., Vocal-cord paralysis and brainstem dysfunction in children with spina bifida. Developmental Medicine and Child Neurology 22: 528-30, 1985.

15. Education and research training

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I have been asked to address the issue of education and research training for rehabilitation professionals (in particular physical and occupational therapists) and the impact of this training on the development of research in these professions. In discussing this issue, I will draw mostly from my personal experience as a physical therapist but will also try to examine the broader context of the non-medical professions. When asked to examine this topic, I interpreted it as a discussion of the potential of present educational systems to promote the quality and quantity of research carried out by rehabilitation specialists. Secondly, I reasoned that an evaluation of the present system should lead to recommendations for improvement. I will divide this discussion into two parts: first, I will comment on the undergraduate educational system and then I will discuss the development of graduate programs in the rehabilitation sciences.

Undergraduate curricula

Before discussing the merits of undergraduate programs, I should state that my belief is that research-training at the undergraduate level should teach critical reasoning, provide enough research methodology and statistical expertise to enable therapists to understand the pertinent literature and also to educate graduates capable of contributing to research projects. This means that I do not expect B.Sc. graduates to become independent researchers. Without making an exhaustive study of the present undergraduate curricula offered to rehabilitation specialists in the different universities across Canada, I am prepared to argue that, on paper at least, these curricula try to promote research interests. For example, the core curriculum which must be followed for accreditation by the Canadian Physiotherapy Association includes objectives related to scientific inquiry which can only be met by courses in research methodology and statistics as well as courses requiring the written or oral communication of research results. I can recall that even in the first B.Sc programs in physical and occupational therapy in the late 60's at McGill University that students were required to do independent library searches of the literature.

The educational approach has, of course, changed dramatically in the last 20 years and this is to be expected of professions which were born at the end of the first world war and really only acquired their specific identities after the second world war. We must remember that the first B.Sc. programs were available in the late 60's, that the first M.Sc. degrees specific to the rehabilitation sciences were offered for the first time in the early 70's in Canada and that the first Ph.D. program specific to the rehabilitation sciences should accept students at McGill University this fall. It is fair to say that the rehabilitation sciences have not attained adulthood as university disciplines recognized by the other more traditional disciplines.

The recognition of the rehabilitation sciences as university disciplines has gradually come about as therapists and medical doctors and other specialists have obtained research degrees and entered the highly competitive world of research. These individuals, who obtained their training in other disciplines and then returned to apply their knowledge to rehabilitation, have gradually changed the education of the professionals and developed the field of research. Physical therapists with Ph.D. degrees obtained in other disciplines (anatomy, neurophysiology, etc) have been teaching in Canadian Universities since the late 70's. Doctoral degrees have also become common among educators in the other disciplines. Since about 1983 a Ph.D. degree is a prerequisite for employment as a regular professor in physical therapy in all 3 Quebec Universities and today a Ph.D. degree or its equivalent is required by all but one of the 13 physical therapy programs across Canada. It has not been possible to limit the recruitment of occupational professors to those with Ph.D. degrees because of the smaller number of occupational therapists who have obtained a Ph.D. In many Universities non-therapists with Ph.D. degrees in closely related fields have been recruited to encourage the development of research and to teach non professional courses. For this reason, specialists in exercise physiology, biomedical engineering, physiology, psychology, kinesiology, physical education, etc. are teaching rehabilitation specialists across Canada.

The increasing number of therapists with doctoral degrees and other research-oriented professors among the Faculty of Rehabilitation programs is changing the emphasis of the teaching. Many basic science courses are now taught by such Faculty who should be able to render the courses more pertinent to rehabilitation while imparting an attitude of scientific inquiry based on experience to the teaching of their courses. A course curriculum can appear excellent on paper but in reality is only as good as the professors who teach the courses. My contention is that research training is best taught by professors who are themselves involved in research and that secondly, a few course credits alone are not sufficient at the undergraduate level. I believe that the whole philosophy of undergraduate programs must be adapted to develop graduates with an acute sense of scientific inquiry.

How can the present undergraduate curricula be improved?

As mentioned above, undergraduate curricula have already undergone major changes over the last 20 years and will no doubt continue to change. In some universities such as Laval University, McGill University and the University of Alberta, the majority of the physiotherapy staff have Ph.D. degrees and an exciting awakening to research has been instilled in the undergraduate program. As more and more research-qualified professors are recruited in the other universities, I predict that the rehabilitation sciences will gradually become like other university disciplines based on a corpus of scientific knowledge. The undergraduate curricula should improve as: 1. more professors have Ph.D. degrees, 2. more professors become involved in research activities, 3. as teaching methods are adapted to change with the philosophy of a curriculum based on scientific inquiry so that "how" becomes less important than "why", and 4. more students are encouraged to partake in research-oriented activities such as special projects during the academic year or summer projects supervised by professors involved in research. Research funds for such projects are available in most universities.

It appears that the Universities in the province of Quebec have revolutionized the teaching of the rehabilitation sciences faster than the universities in the rest of Canada. A major factor in the French-speaking universities, and particularly Laval University, has been the systematic awarding of obligatory study leave for first M.Sc. degrees and then Ph.D. degrees for all non-tenured faculty. Such study leaves were necessary to upgrade the academic qualifications of qualified staff difficult to recruit in general and even more so when language is a barrier. Secondly, the development of rehabilitation technicians in the province of Quebec also accelerated the need to improve University-trained graduates so that the distinction between the two levels was clear. McGill University administrators imposed high academic standards for tenure of educators in physical and occupational therapy in the late 70's and made the development of research a priority. Today, about ten years later, 10 of the 20 professors have doctoral degrees (in 3 years 18 of 22 are expected to have doctoral degrees) and the research base has developed to the point that this group of professors has been judged by external evaluators to be fully capable of offering a doctoral degree in rehabilitation sciences.

The dramatic change that has come about in the province of Quebec suggests that it would be possible to accelerate the process in other provinces as well. Although the smaller population base may in part explain the slower development of research-oriented programs in British Columbia, Saskatchewan, Manitoba and Nova Scotia, it is really not a factor in Ontario. The relative success of programs in Alberta and Quebec is no doubt due to many factors including: provincial government support for the development of research in rehabilitation, individual university commitments to promoting excellence in the field of rehabilitation and of course the all-important factor of leadership. Any of these factors alone cannot be expected to promote success. Consequently, we have to examine the dynamics at the different universities across the country before attempting to pinpoint any specific problem. Another question we have to ask is whether university promotions have hindered the process by granting tenure to staff without higher degrees and who are not engaged in research activities. Such faculty, often resistant to change, can tie up faculty positions for up to 25 years when tenured at about 40 years old. I suggest that the time has come to subject all tenure candidates in the rehabilitation sciences to the same criteria as candidates in other disciplines.

Staff competence and recruitment of faculty

All Universities at present are grappling with problems of staff competence and the recruitment of new Faculty in rehabilitation science programs. The dramatic change in educational standards stimulated in part by the number of therapists obtaining higher degrees has forced the universities to react. Tenured professors at the end of their careers should of course be protected, but all non-tenured staff should be required to upgrade their education to a Ph.D. or its equivalent. Study leaves may be necessary to accelerate this process. In the province of Quebec the second generation of physiotherapy scholars are now applying for teaching positions such that post-doctoral experience has become a prerequisite to obtaining a faculty position. Physical therapists are now also recognized by the funding agencies as research scholars at the same level as scholars in other disciplines.

For at least the next 5 years, and possibly more, university programs in the rehabilitation sciences will face an acute manpower shortage. At present most universities have positions open but cannot find rehabilitation specialists with the required academic credentials to fill them. This means that the pool of expertise is limited and competition for staff positions is not as high as in other disciplines. Furthermore, everyone knows that an academic degree does not guarantee performance. Consequently, study leaves for professors or the hiring of new staff with doctoral degrees cannot be directly equated with success. As the manpower shortage diminishes, natural competition should improve the chances of hiring "the right person". We probably have to also direct our efforts to encouraging the best and the brightest of our undergraduates to pursue graduate degrees and post-doctoral training. This means that we have to offer special grants and incentives to these new graduates because they are attracted to clinical practice and a good salary. Also, undergraduate students are generally high academic performers because of the selection process for admission into rehabilitation programs. Upon graduation, those interested in graduate work or medicine usually have no problem being accepted. Educators in the rehabilitation sciences thus have to offer an interesting alternative to these high achievers so that we can insure the continued development of research in rehabilitation.

Choice of expertise

It used to be that the disciplines of choice for doctoral studies of rehabilitation specialists were: physiology, exercise physiology, neurophysiology, kinesiology, biomechanics, etc. A few chose to obtain doctoral degrees in epidemiology. If we look at the type of research that should advance the practice of rehabilitation, we now realize that at least 3 large categories of research are emerging: 1) the study of pathophysiological processes and changes in these processes with various interventions and 2) the validation of instruments and 3) clinical efficacy studies. Such research interests emphasize the essential role of experts in research methodology and biostatistics. Based on my own experience, I would recommend an expert in research methodology and one in biostatistics for each research unit in rehabilitation. I caution, however, in neglecting the basic sciences or the study of pathophysiological mechanisms underlying the conditions treated in rehabilitation since such studies will further develop the science of rehabilitation. We need a fine balance between both types of researchers. A major weakness in most universities today is that rehabilitation researchers interested in pathophysiological mechanisms do not work with experts in research methodology, possibly because there is a lack of rehabilitation specialists with such expertise. Another weakness is the relatively small number of experts in the psychosocial aspects of the rehabilitation process. Perhaps this need will be met as more occupational therapists chose to study in this area. In summary, I strongly suggest that staff academic upgrading and/or recruitment of new staff should be planned. Not all research units can be experts in all areas. Consequently it may be helpful to plan a concentration of efforts around a central theme for each university. Such a concentration of experts in the basic sciences must be seconded by experts in research methodology and statistics. Perhaps if each university develops a certain area of expertise then exchanges among the universities will be more apt to occur.

Research grants

No matter how you look at it, research costs money; be it for the development of specialized laboratories, the salaries of research assistants, secretarial work, statisticians, the liberation of clinical staff, or even publication costs. The first years of a researcher's career are the most difficult... the start from zero. These first years can be much encouraged by initial "seed" money for laboratory equipment or the initiation of new projects. One must remember that the turnaround time between submission of a grant and notification of the award is about six months, so provisions must be made to financially assist the researcher during this period. Without assistance, a young researcher who must setup a laboratory cannot be expected to reach an appropriately competitive level of function for about 3 years... if of course, funds are obtained on the first try. Financial assistance must be available from the universities. Membership in research groups which obtain funds from provincial, federal or private agencies can also greatly assist a new researcher.

Time allocated to research

Directors of Physical Therapy Programs can promote research activities by providing productive staff members with adequate research time. This may mean that other staff members have to shoulder a greater proportion of the undergraduate teaching load. For example, all tenure-track faculty can be required to engage in quantifiable (by the Director or colleague consensus) research activities (e.g. funded research projects, publications, writing of grant proposals, etc.). Faculty unable or unwilling to engage in such activities can be required to teach more or to assume more committee work or administrative duties. In my opinion, if a new professor is expected to engage in a research career, he must devote at least 40% of his time to research activities. It will be clear to administrators that freeing staff for research activities, especially if all tenured staff are expected to engage in such activities, will result in a dearth of teaching resources. This problem has been countered in some universities by the development of a new status of university teacher, the clinical associate. These persons are rehabilitation specialists with clinical expertise and an M.Sc. degree (whenever possible) who teach courses of a more professional nature. I caution, however, against removing all teaching commitments from research-oriented professors.

Graduate programs

As far as I know, the first M.Sc. program in Rehabilitation Medicine in Canada started at the University of Saskatchewan in 1970 and was followed by a Master of Health Science (Applied) degree at McGill University. These graduate degrees devoted to the training of rehabilitation professionals were of course supplemented by graduate programs in other disciplines which accepted candidates with undergraduate degrees in the rehabilitation sciences. In Canada at the present time, 4 universities with physical and occupational therapy undergraduate programs offer an M.Sc. degree program administered by the professors in the rehabilitation sciences while 2 universities (Laval and Montreal) offer M.Sc. and Ph.D. degrees to students in the rehabilitation sciences within a multidisciplinary graduate program. At the University of Montreal it is a program in Clinical Sciences while at Laval University it is a Neurobiology program. In both universities professors in the rehabilitation sciences supervise graduate students and offer an option in rehabilitation. As previously mentioned, the first Ph.D. degree in Rehabilitation Sciences administered by professors in the rehabilitation sciences should begin this fall at McGill University. Several universities are in the process of creating graduate programs in the rehabilitation sciences.The proliferation of graduate programs, considered further development by the respective universities, leads me to ask whether the assembled faculty in the different universities has sufficient research experience to guarantee a research experience of high calibre to M.Sc. students or to Ph.D. students. It is my belief that we have erred in this respect in the past. In the early 70's it was perhaps necessary to grant the function of supervisor to staff without a Ph.D., its equivalent or even research experience. Can we continue to allow this practice today? Do we impose requirements on our research supervisors such as: publications in the last 2 years, or research funds from an external agency? Teaching graduate students should be a hard-won privilege not the "right" of every university professor.

What type of graduate programs should be developed?

Because of the paucity of qualified personnel, I have been a proponent of basic science multidisciplinary graduate programs. The old adage, "there is safety in numbers" applies to the development of research and graduate programs in the rehabilitation sciences. The worst scenario possible for a new researcher is to face the highly competitive research world alone. Few schools of rehabilitation have the luxury of having 5 - 10 researchers working together in associated fields. For this reason, and for another more subtle reason, that of "instant credibility", I recommend the integration of professors in the rehabilitation sciences into other already established research groups. Such a union provides the new researcher with experienced colleagues willing to help initiate the first projects, to assist in the first grant applications and to generally offer a protective cover until the new researchers can establish a research base and become known to the research community. Let us not kid ourselves, the research world is highly competitive and difficult to break into. Researchers in the rehabilitation sciences need all the help they can get to become recognized as scientists on the same level as those representing other disciplines. Membership of rehabilitation specialists in recognized research groups which are also involved in graduate teaching also insures the cross-fertilization of graduate students, insures assistance in the supervision of the first students and provides graduate students with the classical research model.

Collaboration with therapists in clinical practice

Once graduate programs and research laboratories are firmly established, rehabilitation research faculty can interact with the clinical community to develop research projects. One of the first steps in this process is to implant therapists with M.Sc. degrees in clinical practice. These therapists can then promote the development of research projects and contribute to the formulation and writing of research proposals as well as to the data collection. Rehabilitation faculty can thus engage in collaborative projects and assist clinicians in obtaining research funds. Often a clinician will want to enter the project as a graduate student, thus perpetuating the chances of collaboration.

A Research model in the rehabilitation sciences

An increased awareness for research among clinicians should gradually emerge as a rehabilitation research model is recognized. Already, we have evidence that this process is underway because bright undergraduate students are seeking summer employment in research and continuing onto graduate studies immediately after completion of undergraduate programs. A remarkable change has already occurred in the Quebec universities and at the University of Alberta in the last 10 years. The number of professors with Ph.D.'s and the number of professors engaged in funded research is at least as high and even superior to statistics from more traditional disciplines. For example, $732,711.00 in research funds were awarded to professors in the Faculty of Health Sciences at the University of Alberta for the current year while professors in the School of Physical and Occupational Therapy at McGill University obtained $467,482.00 in 1986-1987 (funds awarded as co-investigators are not included). Furthermore, a large number of physical and occupational therapists are actively involved in the research process as members of internal and external evaluation committees of granting agencies. In the province of Quebec, research in rehabilitation has been given the added impetus of being declared a priority field of study and on a national level, the National Research Council's Associate Committee for Rehabilitation has been actively involved in the promotion of research in rehabilitation for several years.

Summary

My comments on the issue of education and research training have addressed the following points:

- The most important factor for the promotion of good research training is the quality of the professor.

- A research philosophy must permeate undergraduate programs; specific research courses alone are not enough.

- Staff academic upgrading should be encouraged.

- Tenure criteria must be very demanding.

- Recruitment of new staff should be planned to insure a critical mass of researchers in a given field.

- Annexation of rehabilitation researchers with recognized research groups.

- Graduate programs should be developed with caution.

- We must find ways of attracting our brightest undergraduate students to graduate studies.

- The need to develop a recognized research model.

16. The use of correct scientific measurements and analysis in the documentation of physical disability and the extraction of useful diagnostic information.

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The neuromuscular skeletal system is possibly the most complex system to understand, to analyze and to plan therapy. It's complexity arises from the complexity of the central and peripheral nervous system, the number of motoneurons and muscles involved and the number of limbs, joints and degrees of freedom that are being controlled. The total response is the result of a converging of control signals. At the nervous system level each motor unit's activity represents a final common pathway summation of all excitatory and inhibitory inputs to the motoneuron pool. Then at the muscle and joint level there is a second summation of agonistic and antagonistic forces of all active motor units. The moment of force at each joint can be considered as a "final common mechanical pathway". Then at the total limb level we see a further level of integration. Thus the building blocks of the total system result in considerable indeterminacy, which is a nightmare to resolve from a measurement and analysis point of view, but which poses tremendous flexibility and adaptability. When we measure and analyze to tease out information useful for planning therapy we must keep both these points well in our mind.

Valid and reliable measurements and their correct interpretation

If we are to monitor the progress of our patients and are to pinpoint wrong motor patterns we must have confidence in our measurements. I would like to share a few common measurements that are still done incorrectly and therefore are invalid. The first are errors that are introduced in the moment-of-force (torque) measures taken from various isokinetic dynamometers that are now available in many clinics (Winter et al., 1981). Earlier models of dynamometers did not correct for the moment due to gravity. For example, with a patient being assessed for knee extensor strength the machine did not record any moment until the patient had overcome the gravitational load of the leg, foot and lever arm of the machine. Thus the machine read low by an amount which was a function of the load and angle of knee flexion; the percentage error was 30-40% for a moderate contraction, and would likely be higher for patients with below normal quadriceps strength. For knee flexor function gravity assisted the movement, thus the dynamometer reading erred on the high side. Here the error could be even higher because a patient could achieve the knee flexor movement with very little assistance from the knee flexors, most of the moment was due to the gravitational forces alone!

A second area of confusion lies in the area of electromyography that is used in basic research and in clinical applications. In the processing of the raw EMG the term integrated EMG (IEMG) is regularly used. The first use of this term was by Inman and co-workers (1952) when they described a waveform which followed the rise and fall of tension in the muscle. The electronics they employed was a linear envelope detector, not an integrator. Integration is a mathematical term meaning "area under the curve", thus a correct integration of the full-wave rectified EMG would always increase with time. The units of IEMG has been widely abused: everything from mV/s, mV, mV.s to arbitrary units! The correct units for an integrated EMG is mV.s, while the linear envelope is mV. Integration of the EMG over time is a necessary step to calculate its average (in mV) over any given time period. Thus average EMG (AEMG) is a valid term; but to report integrated average is redundant! The common linear envelope processing technique is regularly used to smooth the full wave rectified signal and produce a profile that rises and falls approximately in phase with the tension. The low pass filter used to smooth the EMG introduces a delay, which if chosen correctly has a transfer characteristic (impulse response) that is almost identical to the muscle twitch waveform.

The next two examples are presented not only to demonstrate incorrect measurements and analyses, but also to dramatize some very erroneous interpretations as to what is going on at the motor level in the human body. The first of these deals with the right and wrong way to calculate the net moment-of-force (torque) and the interpretation of the results. This subject has been discussed in detail in one major paper (Wells, 1981) and a letter to the editor (Winter, 1986). The erroneous technique is referred to as the FRFV (Floor Reaction Force Vector) approach and calculates moments as depicted in Figure 1, which shows the ground reaction force vector directed posteriorly during early stance. The FRFV approach calculates each joint moment to be equal to the magnitude of the ground reaction vector times the perpendicular distance between the joint centre and the vector. Thus the ankle moment here would be described as plantarflexor, at the knee it would be flexor and extensor at the hip. The folly of this technique becomes obvious if we extend the vector to calculate the moment of force at the neck. We would estimate the neck moment several times larger than any calculated at the ankle, knee or hip! There are three errors or misinterpretations in this technique.

(i) Numerically, the magnitude of the moment is incorrect because of the mass-acceleration products and moment of inertia-angular acceleration products of the stance limb are not accounted for. We are reminded that the ground reaction vectors represent the algebraic summation of the mass acceleration products of all segments. Using the correct technique those effects must be subtracted as our calculation moves from the ground upwards. Thus by the time we reach the hip joint calculation we have taken into account all inertial forces in the thigh, leg and foot. The FRFV approach does not do this. Numerically the error has been shown to be negligible at the ankle, small but significant at the knee and large at the hip (Wells, 1981).

(ii) The second error in the FRFV technique is the polarity that is attributed to the moment at the joint, and this reflects basic misconceptions about action and reaction. The FRFV is a reaction to the gravitational (external) and muscle (internal) forces acting on the body. As such the reaction force does not cause the moment of force, it merely reflects the cause. An ankle plantarflexor moment, for example, causes a ground action vector to move forward of the ankle joint and this is reflected in the magnitude and the forward location (centre of pressure) of the ground reaction vector. If the ankle muscles were not generating any force the FRFV would remain under the ankle joint.

(iii) final shortcoming of the FRFV approach is that it is not capable of calculating moments during non-weight bearing periods. Such a deficiency is significant in assessment of the swing phase of walking and more so during running.

A second area of rehabilitation where the interpretation of the measurement is quite faulty is the area of postural stability. Force platforms are useful devices for measuring ground reaction forces and also for a direct record of centre of pressure of these reaction forces. In a large number of papers this centre of pressure is described as measuring the sway of the body, which is interpreted as the centre of gravity (C of G) of the body. This is far from the truth. The centre of pressure is not related mathematically to the C of G (Koozekanani et al., 1980) and has been demonstrated experimentally (Shimba, 1984). The centre of pressure (C of P) reflects nothing more than the response of the ankle muscles. In static posture, if the C of P is 5 cms. anterior of the ankle joint and the body weight is 600 Newtons then this means that the ankle plantarflexors are generating 600 x .05 = 30 N.m of torque. The 'action' force of 600 N acting down on the force plate as a result of torque is located 5 cms. in front of the ankle, and the force plate

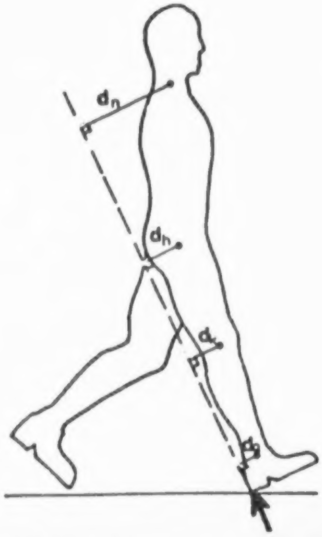

Figure 1
The projection of the ground reaction force vector used to predict the joint moments-of-force. The distance of the projection from the relevant joint center to the vector times the magnitude of that vector gives the net joint moment using FRFV approach (reproduced with permission by R.Wells).

records the magnitude and location of the equal and opposite reaction forces. Figure 2 is presented to demonstrate the differences between C of P and C of G as the body sways back and forth over time. If the action line of C of P and C of G align the body will not be accelerated forwards or backwards. If the C of G lies anterior to the C of P (#1, 4, 5 on Figure 2) the body will be accelerated forwards, if the C of G is posterior to the C of P the acceleration will be backwards (#2 and 3 on Figure 2). At time #1 both the angular velocity and acceleration are forward. At time #2 the plantarflexors increase their activity and the C of P jumps ahead of the C of G and a backward acceleration results in the slowing down of the C of G, but not until time #3 is the body's forward movement arrested and reversed. The body sways backwards and to prevent too much sway in the reverse direction the plantarflexors decrease rapidly (possibly combined with some dorsiflexor activity) so as to slow it down. Then between #4 and #5 the backward sway is reversed and forward sway begins again. Thus to look at the C of P and claim that this is the body sway is incorrect; even if the correct interpretation is made it must be realized that the C of P is the motor response to changes in the C of G. Thus only half the story is present, and without both signals we are unable to assess the motor control system.

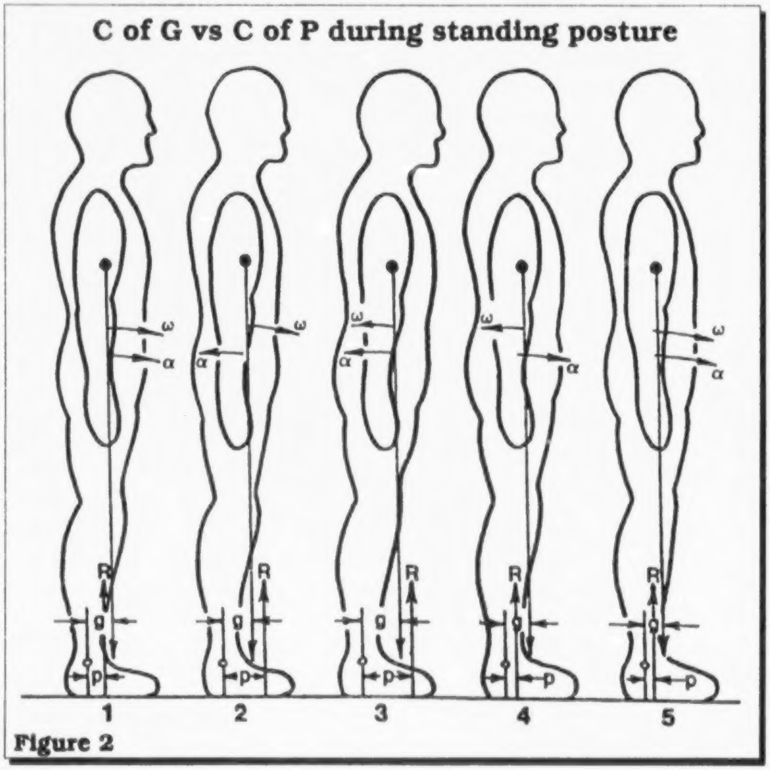

Figure 2

What Measurements to Take?

In the introduction to this paper I dealt with the major problem of complexity and indeterminacy of the neuromuscular skeletal system. In walking this results in tremendous variability in the motor patterns seen at the hip and knee during stance (Winter, 1984) which has been shown not to be random but has been shown to be a very deterministic stride-to-stride adoption to control the dynamic balance of the upper body (Winter, 1987a). Such flexibility is normal and is even more exaggerated in gait pathologies. Thus it is virtually impossible in most gait assessments to predict or hypothesize the changes, and therefore it is difficult to know apriori exactly which of scores of measures to take. We have estimated that close to 100 kinematic and kinetic profile curves plus 20 major EMG's could be affected. Thus we are forced to measure and analyze as much as we can. In other words we use a "shot-gun" approach - we fire straight at the target and maybe get 6 "hits", and more often than not almost all these measures could not have been predicted in advance. I myself have personally challenged the traditional research approach which has developed from sciences where a small number of single measures (Winter, 1987b). Human movement sciences are faced with scores of potentially useful variables, the vast majority vary considerably over the movement period. Thus we are forced to record many measures and to compare them with normal profiles, and with our knowledge of the anatomy and function of a particular movement we can usually pin-point abnormal motor patterns. Thus we are forced to use a shot-gun and we should in no way make apologies for doing so.

References

1. Winter, D.A., Wells, R.P., Orr, G., Errors in the use of isokinetic dynamometers. European J. Appl. Physiol., 46:397-408, 1981.

2. Winter, D.A., Rau, G., Kadefors, R., Broman, H., De Luca, C.J., Units, terms and standards in the reporting of EMG research. Special Scientific Report of the International Society of Electrophysiological Kinesiology, 1980.

3. Wells, R.P., The projection of ground reaction force as a predictor of internal joint moments. Bull, Prosth. Res., 18:15-19, 1981.

4. Bresler, B., Frankel, J.P., The forces and moments in the leg during level walking. Trans.ASEM, 72:27-36, 1950.

5. Winter, D.A., A moment for biomechanics. Letter to editor. Physical Therapy, ??:998-1000, 1986.

6. Inman, V.T., Ralston, H.J., Saunders, J.B., Feinstein, B., Wright, E.W., Relations of human electromyogram to muscular tension. EEG and Clin. Neurophysiol. 4:187-194, 1952.

7. Kozehanani, S.H., Stockwell, C.W., McGhee, R.M., Firoozwand, F., On the role of dynamic models in quantitative posturography. IEEE Trans. Biomed. Eng. BME, 27:605-609, 1980

8. Shimba, T., An estimation of centre of gravity from force platform data. J. Biomech., 17:53-60, 1984

9. Winter, D.A., Kinematic and kinetic patterns in human gait: Variability and compensating effects. Human Movement Science, 3:51-76,1984

10. Winter, D.A., Balance and posture in human walking. Engineering in Med. and Biol., 6:8-11, 1987a

11. Winter, D.A., Are hypotheses really necessary in motor control research? J. Motor Behav., 19:276-279, 1987.

Measurement of physical disability

Comment

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During the discussion period, various issues were addressed including the need for more research and for interdisciplinary collaborative projects. Problems of developing a research team or finding interested people who were able to work together were mentioned. With this in mind I presented a matrix similar to the one below, but incomplete at the time. The idea grew out of a comment a colleague friend made to me some years ago that there were people directly involved in research (R) and people who used or were consumers of research (r). We can also identify the clinician (C) and the consumer of clinical information (c). When looking at the matrix, several types of individuals are identified (the capital letters imply overt action):

	R	r
C	CR	Cr
c	cR	cr

R = The Research Clinician organizes clinical practice using a strict research paradigm and who provides new information and insights to clinical practice.

Cr = This person is a strong clinician open to new ideas, one who applies research in an innovative manner. This person is a clinical practitioner.

cR = This person is primarily a researcher who is aware of clinical problems but does not practice clinically. This person provides basic information to the clinician based upon research.

cr = This person consumes both clinical and research information and does nothing with it, somewhat the obstructionist by not overtly using the information - the antithesis to CR. Another interpretation, but not consistent with the definitions is that cr is the client/patient.

Appendix I

Conference on priorities in childhood physical disability research

List of participants

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KEY

P	Paediatrics
PT	Physical Therapy
OT	Occupational Therapy
CD	Communicative Disorders
O	Orthopaedic Surgery
NS	Neurosurgery
Ep	Epidemiology
N	Nursing
Ed	Education
PS	Plastic Surgery
PSY	Psychology
PSYCH	Psychiatry
E	Engineering
UROL	Urology

Appendix II

Conference program

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PRIORITIES FOR RESEARCH IN CHILDHOOD PHYSICAL DISABILITY

OPENING PLENARY SESSION

AUDITORIUM "A", UNIVERSITY HOSPITAL

Tuesday, 10th May 1988 at 7 p.m.

1. INTRODUCTORY REMARKS AND WELCOME

Howard Keast — Chairman, The Easter Seal Research Institute of Ontario, and Past President, the Easter Seal Society of Ontario

Douglas Reycraft — M.P.P. for Middlesex

Dr. Jacques Robichon — Chairman, Grants Review Committee, Easter Seal Research Institute of Ontario

Dr. James E. Boone — Chairman, Department of Paediatrics, Faculty of Medicine, University of Western Ontario

James Morris — Chairman, Thames Valley Children's Centre

2. FANTASY TIME FOR SCIENTISTS — WHAT I WOULD DO WITH A TEN MILLION DOLLAR RESEARCH GRANT — TOMORROW!

Professor Lennart von Wendt — Goteborg, Sweden

Dr. Lewis Rosenbloom — Liverpool, England

Dr. Martin Bax — London, England

3. ACHIEVEMENTS OF RESEARCH AND DEVELOPMENT — THE CONTRIBUTION OF TECHNOLOGY TO PAEDIATRIC REHABILITATION

Dr. Morris Milner — Hugh MacMillan Medical Centre, Toronto

PRIORITIES FOR RESEARCH IN CHILDHOOD PHYSICAL DISABILITY

Wednesday, May 11 1988

8:45 a.m. **OPENING PLENARY SESSION — SOCIAL SCIENCE AUDITORIUM #2050**

"Tasks and Structure of our Conference"
— Dr. A. Mervyn Fox

"Projections for Physical Disability in Ontario"
— Dr. Gregory Sherman

9:30 a.m. **COFFEE**

10:00 a.m. to 12 noon **THREE CONCURRENT SESSIONS**

1. Session "A" — Room #3010
   Prevention — Reduction of Incidence of Physical Impairment
   Chairperson — Dr. Barry Smith
   Principal Speaker — Dr. Graham Chance
   Discussants — Prof. Lennart von Wendt, Dr. Lewis Rosenbloom
   Secondary Speaker — Dr. Alan German

2. Session "B" — Room #3014
   Reduction of Disability — Communication Technology
   Chairperson — Dr. Morris Milner
   Principal Speakers — Prof. A.F. Newell, Ms. Janice Light
   Discussants — Dr. Cynthia Shewan, Dr. Dennis Harper

3. Session "C" — Room #3018
   Measurement Issues — Measurement of Physical Disability
   Chairperson — Dr. Peter Rosenbaum
   Principal Speaker — Dr. David Winter
   Discussants — Dr. Bruce Gans, Dr. H. deBruin

12 noon to 1:20 p.m. **LUNCH**

1:20 p.m. to 2:45 p.m.

4. Session "D" (PLENARY) — Social Science Auditorium #2050
   Prevention — The Molecular Level
   Chairperson — Dr. Peter Rosenbaum
   Principal Speaker — Priorities for Neuroscience — Dr. N. Lenn
   Discussant — Dr. K. Elisevich
   Secondary Speaker — Priorities for Genetics — Dr. Jack Jung

245 p.m. **TEA**

3:15 p.m. to 5:15 p.m. **TWO CONCURRENT SESSIONS**

5. Session "E" — Room #3010
   Reduction of Disability — Control of the Environment
   Chairperson — Dr. Morris Milner
   Principal Speaker — Dr. S. Naumann
   Discussants — Dr. Charlene Butler, Dr. Carol Ann Parr

6. Session "F" — Room #3014
   Education and Training for Research
   Chairperson — Dr. James E. Boone
   Principal Speaker — Prof. Carol L. Richards
   Discussants — Prof. Martha Piper

PRIORITIES FOR RESEARCH IN CHILDHOOD PHYSICAL DISABILITY

Thursday, 12 May 1988

8:45 a.m.

7. Sesssion "G" (PLENARY) – Social Science Auditorium #2050
Reduction of Severity of Disability – Therapy, Treatment
Chairperson – Dr. Daniel Ling
Principal Speaker – Prof. Martha Piper
Discussants – Dr. B. Gans, Dr. S. Campbell, Dr. C. Butler

10:45 a.m. - 1:15 p.m. **TWO CONCURRENT SESSIONS**

8. Session "H" – Room #3010
Reduction in Disability – Advances in Technology
Chairperson – Mr. Steven Elder
Principal Speaker – Dr. Morris Milner
Discussants – Prof. A.F. Newell, Dr. H. deBruin

9. Session "I" – Room #3014
Measurement Issues – Natural History of Disease: Longitudinal Study
Chairperson – Dr. James E. Boone
Principal Speaker – Prof. Lennart von Wendt
Discussants – Dr. J. McLaughlin, Dr. Lewis Rosenbloom

1:15 p.m. to 2.15 p.m. **LUNCH**

2:30 p.m. to 4:30 p.m. **THREE CONCURRENT SESSIONS**

10. Session "J" – Room #3010
Cerebral Palsy
Chairperson – Dr. Douglas Biggar
Principal Speaker – Dr. Lewis Rosenbloom
Discussants – Prof. Lennart von Wendt, Prof. Martha Piper, Dr. Suzanne Campbell

11. Session "K" – Room #3014
Spinal Dysraphic States
Chairperson – Dr. John F. Schillinger
Principal Speaker – Dr. John Banta
Discussant – Dr. Jeff McLaughlin

12. Session "L" – Room #3018
Reduction of Handicap
Chairperson – Dr. Ben Goldberg
Principal Speaker – Dr. Dennis Harper
Discussants – Dr. David Cadman, Dr. Naomi Rae-Grant

PRIORITIES FOR RESEARCH IN CHILDHOOD PHYSICAL DISABILITY

Friday, 13th May 1988

COLLOQUIUM

"THE TRIBULATIONS OF RESEARCH FUNDING"

10:00 a.m. to 12 noon

Room #210 Somerville House

The University of Western Ontario

Moderator — Dr. Bessie Borwein
Assistant Dean, Research, Faculty of Medicine,
University of Western Ontario

Panel — Mr. Ernest Jodoin
Vice President, The Hospital for Sick Children Foundation

— Dr. Ron Heacock
Director, National Health Research Development Program, Ottawa

— Ms. Pat Stuckless
Policy Development and Research Division, Ontario Ministry of Health

— Dr. Robert Colcleugh
Associate Professor, Surgery, University of Western Ontario

— Ms. Ruth Koheil
Research Coordinator, The Hugh MacMillan Medical Centre, Toronto

Kids with physical disabilities are getting too much attention.

Nobody likes to be stared at. Or pointed at. But when you have a disability, that's the kind of thing that happens to you every day. And the worst part is, after awhile, you start believing that there really is something wrong with you. It's time to change that. And start paying more attention to the person. Not the disability.

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